JIDR Journal of Intellectual Disability Research
Published on behalf of mencap and in association with IASSID

Journal of Intellectual Disability Research doi: 10.1111/jir.12827

VOLUME 65 PART 6 pp 501–534 JUNE 2021

Systematic Review

Mental ill-health in mothers of people with intellectual disabilities compared with mothers of typically developing people: a systematic review and meta-analysis

E. Rydzewska, K. Dunn, S.-A. Cooper & D. Kinnear

Institute of Health and Wellbeing, College of Medical, Veterinary and Life Sciences, University of Glasgow, Glasgow, UK

Abstract

Background Mothers of people with intellectual disabilities (IDs) face exceptional challenges and may be more prone to experiencing mental ill-health compared with mothers of typically developing people. These mental ill-health problems may differ at different stages of the caregiving trajectory. However, there is no evidence synthesis on this topic. We aimed to systematically review evidence in this area and identify gaps in the existing literature.

Method Prospero registration: CRD42018088197. Medline, Embase, CINAHL and PsycINFO databases were searched. No time limits were applied. Studies were limited to English language. Inclusion criteria were studies of mothers of people with IDs that also included a comparison group of mothers of typically developing/developed children. Data were extracted from selected studies using a structured database. Study selection and quality appraisal were double rated. Where possible, meta-analyses were performed.

Results Of the retrieved articles, 32/3089 were included, of which 10 reported on anxiety, 21 on depression and 23 on other indicators of mental ill-health. Overall, previous studies reported that mothers of people with IDs experienced poorer mental health as compared with mothers of typically developing people. Meta-analyses revealed significant findings for anxiety, depression, parenting stress, emotional burden and common mental disorders, but not for somatic symptoms. However, there was a considerable heterogeneity; hence, interpretation of results should be cautious. Identified gaps included scarce research on mental ill-health of mothers of adults with IDs at different stages of the caregiving trajectory.

Conclusions There is evidence of poorer mental ill-health in mothers of people with IDs compared with mothers of typically developing people, but lack of focus on different stages of the caregiving trajectory, methodological inconsistencies between studies and lack of robust studies pose limitations. This highlights the need both for improved support for mothers of people with IDs and for further methodologically robust research.

Keywords anxiety, depression, intellectual disabilities, mental health, mothers, systematic review

Correspondence: Dr Deborah Kinnear, Institute of Health and Wellbeing College of Medical, Veterinary and Life Sciences Mental Health and Wellbeing, University of Glasgow, 2nd Floor, Admin Building Gartnavel Royal Hospital 1055 Great Western Road Glasgow, G12 0XH. Tel. +44 (0) 141 211 0688. (e-mail: deborah.kinnear@glasgow.ac.uk).

Background

Reviews of evidence on prevalence of mental ill-health

Mothers of children with intellectual disabilities (IDs) have been reported to have poorer mental health compared with other carer groups (e.g. those supporting a child with physical disabilities or an elderly parent with dementia) (Pinquart and Sörensen 2003). These mothers also appear to have more depressive, anxiety and stress symptoms than fathers of children with IDs (Singer 2006; Montes and Halterman 2007; Cairns *et al.* 2012, 2014; Giallo *et al.* 2015). However, most studies include small and highly selective samples, without comparison groups of parents of typically developing children (e.g. Chen *et al.* 2001; Azeem *et al.* 2013; Dawson *et al.* 2016; Rimmerman *et al.* 2018). We believe that research including large samples with comparison groups of mothers of typically developing people is important to undertake in view of health inequalities potentially associated with IDs. Should such associations be found, there would be implications for family systems and child development, particularly given the potential to then advocate for early preventative interventions.

A brief narrative overview of studies on mental health needs of parents with IDs reported a high prevalence of mental health needs in this population. However, the review was not systematic, and included studies without comparison groups of mothers of typically developing children (O'Keeffe and O'Hara 2008). A further systematic review found an association between positive impact of caring for a child with IDs and wellbeing in parents of children with IDs, with poor wellbeing accompanied by lower positive perceptions and higher negative perceptions of caregiving. However, this relationship was affected by extraneous variables including partner wellbeing, challenging behaviour, and syndrome characteristics (Horsley and Oliver 2015).

A research critique (Bailey *et al.* 2007) and two meta-analyses (Singer 2006; Miodrag *et al.* 2015) on maternal depression and stress in families of children with various disabilities or chronic health conditions reported that mothers of children with disabilities exhibited higher rates of depressive symptoms, stress and clinical levels of depression. A literature review of studies on stress, poor sleep and wellbeing in mothers of children with developmental disabilities found that they experienced more of these problems than mothers of typically developing children, and these problems were more persistent (Lee 2013). We did not locate any systematic reviews investigating prevalence of mental ill-health in mothers of people with IDs compared with mothers of typically developing children.

Mental ill-health at different stages of the caregiving trajectory

Improvements in health and social care and deinstitutionalisation mean that more people with IDs are living in the community and enjoying longer lives (Graham *et al.* 2013). Due to this prolonged caregiving role, parent carers of people with IDs face exceptional challenges and have very different circumstances from those people who come to care for relatives following onset of illness or trauma-related disability (Innes *et al.* 2012). Caregivers of children with IDs need more information on education and therapy (Jackson *et al.* 2016) in addition to help with parenting and coping (Douma *et al.* 2006; James 2013), especially if their child also experiences complex healthcare needs (Tan 2017). All of these factors might impact on mothers' mental health in different ways at different stages of the caregiving trajectory (i.e. caring for a child, adult and older adult) (Emerson and Hatton 2011), but this is less clearly understood in the research. While caring can be an extremely positive and rewarding experience (Scorgie and Sobsey 2000; Jokinen and Brown 2005), small-scale studies suggest it may also impact on parents' mental health at key points in the caregiving trajectory (e.g. Chen *et al.* 2001; Cairns *et al.* 2014). Some research has directly compared mental health impacts on mothers at different ages, but has yet to compare impact on mothers with differently aged children. Chen *et al.* (2001) carried out a survey in the US looking at the health of 108 mothers of adults with IDs, divided into mid-life (younger) groups of women between 55 and 64 years of age and later-life (older) groups aged 65 years and over. The study found that caring for adult children with IDs might not be as detrimental to the mid-life mothers' physical component of health as it might be to the later-life mothers. Both groups reported self-reported physical and mental health

scores similar to the US national norms. Cairns *et al.* (2014) carried out a survey with 100 older parent carers subgrouped into young-old (65–74 years), middle-old (75–84 years) and old-old (85 + years) parent carers and found that mean mental health scores for the young-old (65–74) and middle-old (75–84) parent carers were below 50, that is, below the UK general population norms. While parent carers in the old-old (85+) group scored just above 50 (i.e. above average) compared with the two younger subgroups, the mean score for all three subgroups combined was still below UK norms. We did not locate any systematic reviews investigating prevalence of mental ill-health in mothers of people with IDs at different stages of the caregiving trajectory.

For these reasons, we aimed to systematically review mental ill-health in mothers of people with IDs compared with mothers of typically developing/developed children and adults and investigate impact of caring on mental ill-health at different stages of the caregiving trajectory. Specific research questions were as follows:

1 How common is mental ill-health in mothers of people with IDs compared with mothers of typically developing people, and at different stages of the caregiving trajectory?
2 Are there gaps in the evidence base on mental ill-health in mothers of people with IDs compared with mothers of typically developing people, and at different stages of the caregiving trajectory?

Methods

This review was prospectively registered with the International Prospective Register of Systematic Reviews (PROSPERO, registration number: CRD42018088197). The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist, flow diagram and guidelines were followed (Shamseer *et al.* 2015). Four databases were searched: Medline, Embase, CINAHL and PsycINFO in February 2020. Search terms included terms to capture IDs, combined with terms on mental health and parent carers (detailed list is included in Appendix 1). Studies were limited to English language. No time limits were applied.

Study screening and selection

Papers were initially screened on their title and abstract by the first reviewer (E. R.). A second reviewer (K. D.) read a random 10% of titles and abstracts to ensure the selection approach was systematic. Both reviewers were post-doctoral researchers. Any discrepancies were planned to be resolved through discussion, with further double reviewing planned if consistency was not reached by this stage. Relevant full texts were then reviewed for eligibility by the first reviewer (E. R.). For four papers for which there were doubts on inclusion, papers were read by two more reviewers (K. D. and D. K.) and were subsequently excluded from the data extraction stage. Data were then extracted from the selected papers by the first reviewer (E. R.), using a structured database, on author; publication year; country; population size and type; comparison group; definition of mental ill-health; measures used; findings; and study quality. Additionally, we reviewed the reference lists of all included studies to check if they referenced any relevant publications that were not identified through our electronic searches. Strict inclusion and exclusion criteria were used.

Inclusion criteria

- Mother caring for a child with IDs (IQ ≤ 70, or children described as having 'intellectual disability', 'mental retardation' or equivalent terms listed in our search terms);
- Mothers aged 16 years and above;
- Observational studies, for example, cohort, case–control and cross-sectional studies;
- Studies from peer-reviewed journals.

Exclusion criteria

- Data on mothers of children with IDs were not separately reported and were less than 50% of the sample;
- Studies without a comparison group of mothers of typically developing children;
- Syndrome specific studies (e.g. Down syndrome, Williams syndrome, Fragile X syndrome and autism);
- Studies on samples with developmental delay;
- Non-human studies;
- Grey literature.

Quality assessment

The 12-item Critical Appraisal Skills Programme Checklist (CASP 2017) for cohort studies was used to systematically assess study quality. This covers all domains of quality assessment of observational studies (Sanderson *et al.* 2007), including clarity of the stated aims, methodology (including age/gender standardisation and consideration of group differences in disease prevalence rates), study design and size, participant selection, measures used, data collected, analyses employed, results, bias, generalisability, conflict of interests, and ethical procedures. Quality appraisal was double rated. First reviewer (E. R., research fellow) rated all papers while the second (K. D., research associate) and third (D. K., senior lecturer) reviewer second rated half of the reviewed papers each. Freedom from risk of bias was appraised per domain: 'Yes' (low risk), 'Cannot tell' (unknown risk) or 'No' (high risk). The following classification was used for risk of bias for each study overall (Mathie *et al.* 2017):

- Rating A = low risk of bias for all 12 items;
- Rating B*x* = uncertain risk of bias for *x* items, low risk of bias in all other items;
- Rating C*y*,*x* = high risk of bias in *y* items, uncertain risk of bias in *x* items and low risk of bias in all other items.

Meta-analysis of studies

Meta-analysis was undertaken using Review Manager (version 5.4). All outcome measures from the included studies were reported on a continuous scale. Only studies that reported on the mean, standard deviation and sample size for mothers of people with IDs and the comparison groups were included in the meta-analyses, as these statistics were necessary to calculate an unbiased standardised mean difference (SMD) between the studied populations. A negative mean difference indicated poorer mental health for the comparison group than for mothers of people with IDs. For papers that included more than one group of mothers of children with IDs (e.g. mothers of children with IDs and autism), but only one comparison group for typically developing population, we calculated a combined mean and standard deviation for these subgroups to minimise bias. Effect size was interpreted as small when SMD < 0.40, moderate when SMD ranged from 0.40 to 0.70 and large when SMD > 0.70 (Schünemann *et al.* 2011).

The Chi-squared statistic I^2 was chosen to measure the level of heterogeneity across the studies, as it allows for interpretation of results regardless of the number of studies included in the meta-analysis or their type of outcome data or effect measurement (Higgins *et al.* 2003). Heterogeneity was interpreted as not observed when $I^2 = 0\%$, low when $I^2 = 25\%$, medium when $I^2 = 50\%$ and high when $I^2 = 75\%$ (Higgins *et al.* 2003). Random effects models were selected for this analysis due to the different measures (e.g. different indicators of mental health) in the included studies. We intended to assess publication bias using funnel plot techniques and Egger's regression test as appropriate, given the known limitations of these methods. The meta-analysis was performed on different subgroups, including anxiety, depression and other indicators of mental ill-health.

Sensitivity analysis

Sensitivity analysis was used to assess the impact of risk of bias for each study on the pooled SMD. Data were removed one-by-one from the meta-analysis for each study, beginning with the lowest ranked papers, to determine the effect of each individual study on the pooled SMD.

Results

The search returned 4613 records, of which 251 articles were read in full and 32 selected for inclusion and data extraction (Fig. 1). There were no disagreements between reviewers ($\kappa = 1.00$). Of the 32 studies, 16 had appropriate data for meta-analysis, although all meta-analyses revealed high heterogeneity, and hence, the meta-analysis results must be treated with considerable caution.

Table 1 presents main study characteristics. Four were from the UK, 10 from the USA, six from Sweden, two from Australia, two from Iran and eight from other countries. Twenty-six were from high-income countries (UK, USA, Australia, Sweden, Germany, Italy, Israel and Bahrain), four from upper middle-income countries (Iran, Malaysia and Turkey) and two in lower middle-income

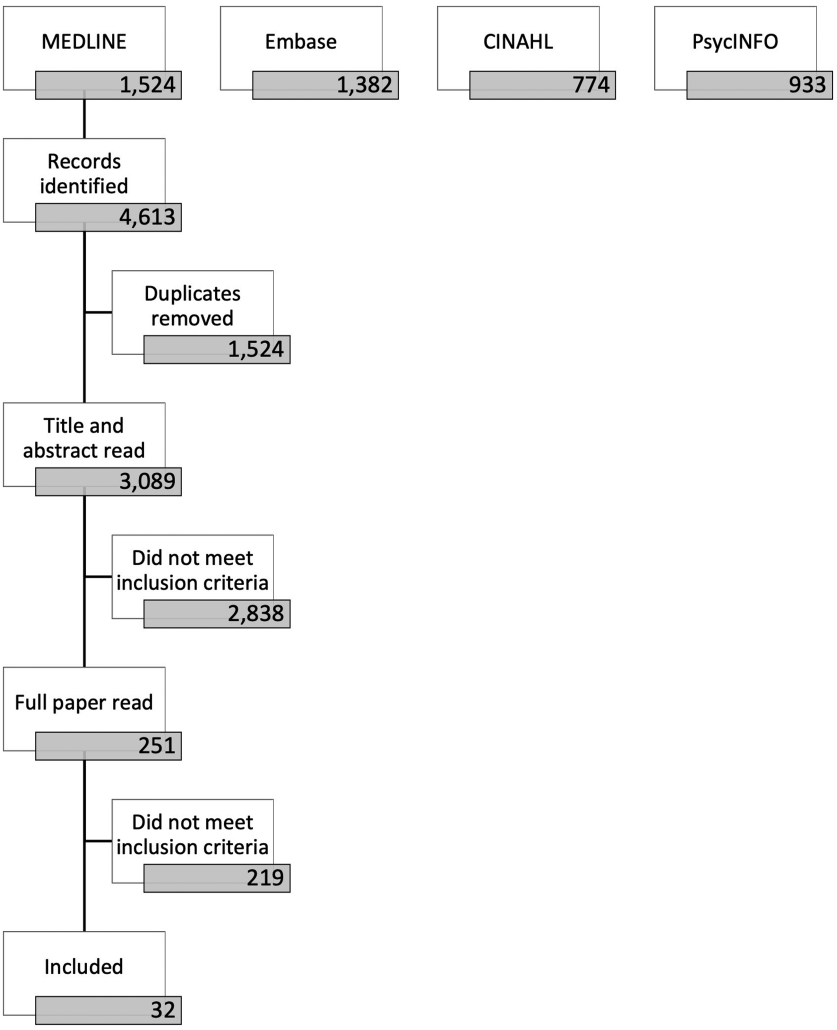

Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart of the study selection process

countries (Nigeria and India) (World Bank 2020). Publication dates were 1966–2019, with seven studies published before 2000. The majority included mothers whose children were under the age of 18 (n = 29); only two included mothers of both children and adults (both by the same author using the same Australian dataset), and one included mothers of adults only. Four out of 32 studies used unvalidated measures; a 46-item sentence completion test constructed by Cummings *et al.* (1966), self-rated health scale (Olsson and Hwang 2008), a 4-point scale on a self-completion questionnaire (Emerson *et al.* 2006) and a questionnaire from an unpublished preliminary study (Lenhard *et al.* 2005). A further two used ICD-9 and ICD-10 codes that had been recorded in health registries (Fairthorne *et al.* 2015; Fairthorne *et al.* 2016). Publication bias was not assessed as there were inadequate numbers of included studies to properly assess a funnel plot or conduct more advanced regression-based assessments.

We did not exclude papers based on quality, but quality appraisal revealed limitations to most of the studies. Small and selective samples, lack of precise reporting of findings or confounding factors in the design and analysis were the most common flaws (further details included in Appendix 2). Eight papers had low risk of bias, 12 had uncertain risk of bias and 12 presented high risk of bias (Table 1). There was a near perfect agreement between reviewers (κ = 0.95).

Table 1 Characteristics of reviewed studies

Year	Author	Country	Mothers ID (N)	Mothers ID Age (years)	Mothers TD (N)	Mothers TD Age (years)	Mothers recruited from	Risk of bias	Risk of bias rating
Studies of mothers of children									
1966	Cummings *et al.*	USA	60	–	60	–	Clinics, social agencies and community physicians	Low	A
1978	Miller & Keirn	USA	44	*M* = 34.8	41	*M* = 36.7	University neuropsychiatric outpatient clinic and local newspaper/telephone recruitment	High	C2.1
1989	Harris & McHale	USA	30	*M* = 34.52, SD = 7.93	30	*M* = 36.28, SD = 3.61	Schools, social and medical services	Uncertain	B2
1989	Walker *et al.*	USA	24	–	24	–	Special education schools and general paediatrics clinics records	Uncertain	B1
1992	Walker *et al.*	USA	24	–	24	–	Special education schools and general paediatrics clinics records	High	C2.1
1993	Andersson	Sweden	47	*M* = 34	47	*M* = 34	Disability services and personal contacts	High	C2
1999	Ong *et al.*	Malaysia	75	*M* = 36.3, SD = 6.1	75	*M* = 37.1, SD = 6.6	Paediatric hospital institute and outpatient clinic	Uncertain	B1
2001	Olsson & Hwang[†]	Sweden	145	–	204	–	Disability services and randomly selected control families living in the same area	Low	A
2002	Olsson & Hwang[†]	Sweden	144	–	202	–	Disability services and randomly selected control families living in the same area	Uncertain	B1
2002	Weiss	USA	40	24–48 years old	40	–	Disability services, statewide developmental disabilities conference and parent networking organisation	Low	A
2003	Emerson	UK	245	–	9481	–	Secondary data analysis of the ONS survey of the Mental Health of Children and Adolescents in Great Britain, 1999	High	C2.1
2005	Eisenhower *et al.*[†]	USA	43	*M* = 36.0	136	*M* = 35.0	Disability services, preschools and day care centres	Uncertain	B1
2005	Lenhard *et al.*[†]	Germany	66	Age at child's birth: *M* = 28.2, SD = 4.9	69	Age at child's birth: *M* = 31.5, SD = 4.3	Special education and mainstream schools	High	C1.3
2006	Abasiubong *et al.*	Nigeria	106	*M* = 40.0	101	*M* = 34.8	Disability services and higher education institution	High	C1.1

Table 1. *(Continued)*

Year	Author	Country	Mothers ID (N)	Mothers ID Age (years)	Mothers TD (N)	Mothers TD Age (years)	Mothers recruited from	Risk of bias	Risk of bias rating
2006	Emerson *et al.*	UK	514	–	6440	–	Secondary analysis of the Department for Work and Pensions' Families & Children Study, Wave 4	High	C2.2
2006	Olsson & Hwang	Sweden	167	*M* = 39.8	185	*M* = 39.3	Disability services and randomly selected control families living in the same area	Uncertain	B2
2007	Mugno *et al.*[†]	Italy	49	–	42	–	Neurological and psychiatric centre, mainstream schools	Low	A
2008	Neece & Baker	USA	74	*M* = 36.1, SD = 6.5	115	*M* = 37.9, SD = 5.9	Community services for people with developmental disabilities, preschools and daycare programmes	Uncertain	B1
2008	Olsson & Hwang	Sweden	62	–	178	–	Disability services and SPAR register of all individuals living in Sweden	Uncertain	B2
2010	Gupta & Kaur	India	30	–	13	–	Special education and mainstream schools	High	C3
2011	Mirsaleh *et al.*	Iran	124	25–50 years old	124	25–50 years old	Special education and mainstream schools in southeastern Tehran	High	C2
2011a	Totsika *et al.*[†]	UK	412	–	14 444	–	Secondary data analysis of the Millennium Cohort Study	Uncertain	B1
2011b	Totsika *et al*[†]	UK	590	>70% 30–44 years old	17 727	>70% 30–44 years old	Secondary analysis of two UK national surveys on psychiatric morbidity of 5–16-year-olds	Uncertain	B2
2013	Kilic *et al.*	Turkey	Special education: 75; Mainstream education: 70	23–29 years *N* = 55; 30–36 years *N* = 48; 37 + years *N* = 42	75	23–29 years *N* = 29; 30–36 years *N* = 16; 37 + years *N* = 30	Disability services	High	C1.1
2013	Norlin & Broberg	Sweden	T1: 58; T2: 46	*M* = 34.0	T1: 178, T2: 131	*M* = 33.8	Disability services and SPAR register of all individuals living in Sweden	Low	A
2015	Long *et al.*	USA	Latina: 48; non-Latina: 50	Latina: *M* = 36.58, SD = 5.61; non-Latina: *M* = 42.02, SD = 5.18	Latina: 49; non-Latina: 45	Latina: *M* = 38.68, SD = 6.00; non-Latina: *M* = 39.78, SD = 6.52	Hospital-based general and specialty paediatric programmes, community agencies, paediatricians' offices, public and private schools, and word of mouth	Uncertain	B1
2018	AlAnsari and Jahrami	Bahrain	30	*M* = 33.3, SD = 6.5	30	*M* = 32.4, SD = 4.4	National intellectual disability/autism registry and social contacts of mothers in ID group	High	C2.1

Table 1. *(Continued)*

Year	Author	Country	Mothers ID (*N*)	Mothers ID Age (years)	Mothers TD (*N*)	Mothers TD Age (years)	Mothers recruited from	Risk of bias	Risk of bias rating
2018	Hosseinink *et al.*	Iran	Conflicting sample sizes (94, 40 and 48) reported in the abstract, methods and tables	–	Conflicting sample sizes (94, 40 and 48) reported in the abstract, methods and tables	–	Special education schools and mothers of TD children residing in Yasouj city	High	C5.2
2019	Blacher & Baker[†]	USA	28	*M* = 43.3	84	*M* = 45.5	Diagnostic/intervention services for people with developmental disabilities, preschools, schools and daycare programmes	Low	A
Studies of mothers of children and adults									
2015	Fairthorne *et al.*[†]	Australia	Mild–moderate ID: 4629; Severe ID: 322	Age at index birth: mild–moderate ID: <20 years *N* = 519; 20–34 years *N* = 3668; >35 years *N* = 492; severe ID: <20 years *N* = 24; 20–34 years *N* = 249; >35 years *N* = 49	271 249	Age at index birth: <20 years *N* = 19 764; 20–34 years *N* = 221229; >35 years *N* = 32 256	Population of women who gave birth to a live child in Western Australia between Jan 1983 and Dec 2005	Low	A
2016	Fairthorne *et al.*[†]	Australia	5927	Age at index birth: <20 years *N* = 637; 20–34 years *N* = 4687; >35 years *N* = 603	271 249	Age at index birth: <20 years *N* = 22 085; 20–34 years *N* = 235 820; >35 years *N* = 32 679	Population of women who gave birth to a live child in Western Australia between Jan 1983 and Dec 2005	Low	A
Studies of mothers of adults									
2001	Rimmerman & Muraver	Israel	160	*M* = 68.6	128	*M* = 69.6	Sheltered employment programme	Uncertain	B1

Abbreviations: DS, Down syndrome; ID, children with intellectual disability; *M*, mean; SD, standard deviation; TD, typically developing children.
[†]These papers additionally separately analysed subgroups of people with intellectual disabilities and Down syndrome and/or intellectual disabilities and autism.

Table 2 Studies investigating anxiety in mothers of children with intellectual disabilities

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
Studies of mothers of children						
1978	Miller & Keirn	*M* = 52.63, SD = 8.11	*M* = 52.35, SD = 7.24	Not reported	MMPI	elevations reflect parents' pathological reactions to the presence of a disabled child; psychasthenia scale
1993	Andersson	Short experience: *M* = 7.03; long experience: *M* = 6.72; total: *M* = 6.92	Short experience: *M* = 7.14; long experience: *M* = 6.71; total: *M* = 6.98	No statistically significant differences found between the groups	HADS	HADS: anxiety score ≥8
2002	Weiss	*M* = 14.4, SD = 2.82	*M* = 13.2, SD = 2.65	$F = 3.45$, $P = .035$	Questionnaire developed by the US Department of Health, Education and Welfare	7 questions on anxiety-related (e.g. jitteriness, nervousness, irritability, anger) symptoms
2003	Emerson	36.0% worried to a great extent; 34.0% to some extent	5.0% worried to a great extent; 15.0% to some extent	Mann–Whitney $z = 19.61$, $P < .001$	SDQ	Self-assessed psychological impact of the difficulties of the sampled child/adolescent on the respondent
2005	Lenhard *et al.*	51.9% had a significantly elevated score	31.8% had a significantly elevated score	Significantly elevated score in ID mothers compared with TD mothers ($P = .004$)	STAI	Not reported
2006	Abasiubong *et al.*	*N* = 27 (25.5%) with score ≥8	*N* = 7 (6.9%) with score ≥8	Not reported	HADS	HADS: anxiety score ≥8
2011	Mirsaleh *et al.*	*M* = 7.88, SD = 4.62	*M* = 5.56, SD = 2.79	$t = 4.79$, $P < .001$	GHQ-28	GHQ-28: symptoms of anxiety/insomnia; higher score indicates worse health
2013	Kilic *et al.*	Special education X = 52.79, SS = 8.7; mainstream education X = 56.73, SS = 13.7	X = 49.87, SS = 6.5	$F = 8.579$, $P < .001$	STAI	STAI: score > 60
2018	AlAnsari and Jahrami	*M* = 2.43, SD = 0.4	*M* = 1.03, SD = 0.27	$P = .008$	GHQ-28	GHQ-28: symptoms of anxiety/insomnia; higher score indicates worse health
Studies of mothers of children and adults						
2015	Fairthorne *et al.*	Neurotic disorders: mild–moderate ID: 326 (7.0%); severe ID: 19 (5.9%)	Neurotic disorders: 8038 (3.0%)	Mild–moderate ID: IRR = 1.80, 95% CI [1.3, 2.5]; severe ID: IRR = 1.98, 95% CI [0.6, 6.4]	ICD-9 and ICD-10 codes	Statutory state-based registries of hospital admissions and outpatient mental health appointments

Abbreviations: CES-D, Centre for Epidemiological Studies Depression Scale; DIGS, Diagnostic Interview for Genetic Studies; GHQ, General Health Questionnaire; HADS, Hospital Anxiety and Depression Scale; ID, children with intellectual disability; *M*, mean score; MMPI, Minnesota Multiphasic Personality Inventory; SD, standard deviation; SS, sum of squared deviations; STAI, State–Trait Anxiety Inventory; TD, typically developing children.

Anxiety

Ten studies investigated anxiety (Table 2); nine on mothers of children and one on mothers of children and adults. Two studies used each of the following measures: Hospital Anxiety and Depression Scale (Andersson 1993; Abasiubong *et al.* 2006), State–Trait Anxiety Inventory (Lenhard *et al.* 2005; Kilic *et al.* 2013) and General Health Questionnaire, which has an anxiety sub-scale (Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018). The remaining four studies used a variety of other instruments (Miller and Keirn 1978; Weiss 2002; Emerson 2003; Fairthorne *et al.* 2015).

All studies apart from two (Miller and Keirn 1978; Andersson 1993) reported more anxiety in mothers of children with IDs. The biggest differences were observed by Abasiubong *et al.* (2006), Emerson (2003), and Lenhard *et al.* (2005). Rates of anxiety in the secondary analysis of routinely collected administrative data on mothers of both children and adults (Fairthorne *et al.* 2015) were lower than in some studies of mothers of children only (e.g. Lenhard *et al.* 2005), but direct comparisons cannot be made due to differing methodological approaches.

Data were suitable for meta-analysis in four studies (Miller and Keirn 1978; Weiss 2002; Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018) (Fig. 2). The pooled SMD for differences in anxiety levels between mothers of children with IDs and mothers of typically developing children was statistically significant at 1.18, 95% confidence interval (CI) [0.18, 2.17], $P < .05$. There was a large effect size, demonstrating that mothers of children with IDs experience heightened anxiety compared with mothers of typically developing children. Heterogeneity between studies was, however, at a high level, with $I^2 = 95\%$, and the significant finding was largely driven by the study by AlAnsari and Jahrami (2018).

Depression

Twenty-one studies investigated depression (Table 3); 20 studies on mothers of children and one on mothers of children and adults. Eight used the Beck Depression Inventory (Harris and McHale 1989; Kilic *et al.* 2013; Norlin and Broberg 2013; Olsson and Hwang 2001, 2002, 2006, 2008; Weiss 2002), two used the Centre for Epidemiological Studies Depression Scale (Walker *et al.* 1989; Eisenhower *et al.* 2005), Hospital Anxiety and Depression Scale (Andersson 1993; Abasiubong *et al.* 2006) and General Health Questionnaire, which has a depression sub-scale (Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018). Seven remaining papers used a variety of other instruments (Cummings *et al.* 1966; Miller and Keirn 1978; Ong *et al.* 1999; Emerson 2003; Emerson *et al.* 2006; Fairthorne *et al.* 2015; Long *et al.* 2015).

All studies reported higher depression scores in mothers of children with IDs, except one reporting statistically non-significant lower scores in mothers of children with IDs (Walker *et al.* 1989). When Emerson *et al.* (2006) controlled for maternal characteristics (i.e. age, marital status, general health status and health problems limiting activity), the association between caring for a child with IDs and maternal happiness was no longer statistically significant.

Data were suitable for meta-analysis in 13 studies (AlAnsari and Jahrami 2018; Cummings *et al.* 1966; Eisenhower *et al.* 2005; Harris and McHale 1989; Miller and Keirn 1978; Mirsaleh *et al.* 2011; Norlin and Broberg 2013; Olsson and Hwang 2002, 2006, 2008; Ong *et al.* 1999; Walker *et al.* 1989; Weiss 2002) (Fig. 3). Norlin and Broberg (2013) collected data at two time points. Due to a decrease in number of participants at time 2, we meta-analysed data for time

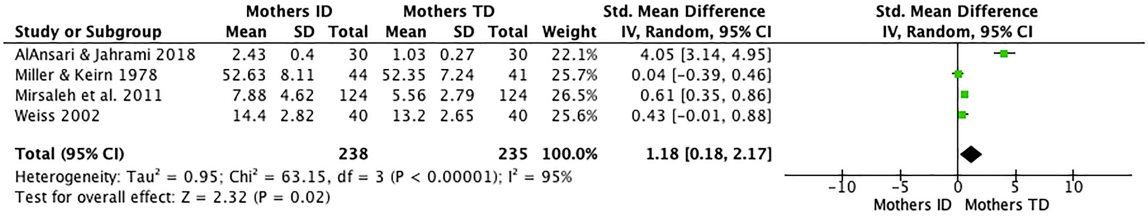

Figure 2. Forest plot for four analysable studies presenting findings on anxiety in mothers of people with intellectual disabilities. Shows standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]

Table 3 Studies investigating depression in mothers of children with intellectual disabilities

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
Studies of mothers of children						
1966	Cummings *et al.*	*M* = 7.3, SD = 2.0	*M* = 5.3, SD = 2.1	$t = 5.26$, $P = .01$	Bespoke Sentence Completion Test	Level of depressive feeling, higher score indicates higher level
1978	Miller & Keirn	*M* = 56.23, SD = 10.33	*M* = 53.24, SD = 7.30	Not reported	MMPI	Elevations reflect parents' pathological reactions to the presence of a disabled child; depression scale
1989	Harris & McHale	*M* = 5.77, SD = 5.00	*M* = 3.87, SD = 4.24	No significant group differences, but ID mothers reported somewhat higher depression; the level of symptomology ranged from no depression to mild depression	BDI	Score range of 0–63, with higher scores representing more intense depressive symptomology
1989	Walker *et al.*	*M* = 9.67, SD = 9.75	*M* = 10.12, SD = 10.08	$F_{3,\ 91} = 0.39$, P not statistically significant; ID: $\beta = -.03$ ($B = -.73$), P not reported	CES-D	CES-D questionnaire to measure depressive symptoms in adults, 20 items, 4-point scale, score range 0–60, score > 16 for depression
1993	Andersson	*M* = 4.30	*M* = 3.60	No statistically significant differences found between the groups	HADS	HADS: depression score ≥8
1999	Ong *et al.*	*M* = 24.7, SD = 5.8	*M* = 21.5, SD = 3.5	$P < .001$	PSI	Child domain (CDS) and parent-domain stress (PDS), Life Stress and a total PSI score
2001	Olsson & Hwang	*M* = 9.2, dysphoria *N* = 53 (37.0%), depression	*M* = 5.2, dysphoria	Not reported	BDI	BDI: 4-point scale of 0–4 with score 10–20 for dysphoria and >20 for depression
2002	Olsson & Hwang	*N* = 12 (8.0%) 1. *M* = 9.2, SD = 7.4 2. Low SoC *M* = 12.1 High SoC *M* = 4.5 Dysphoria low SoC *N* = 44 (50.5%) Dysphoria high SoC *N* = 9 (16%) Depression low SoC *N* = 12 (14.0%) Depression high SoC *N* = 0 (0%)	*N* = 27 (13.0%), depression *N* = 7 (4.0%) 1. *M* = 5.2, SD = 5.2 2. Low SoC *M* = 9.7 High SoC *M* = 2.3 Dysphoria low SoC *N* = 23 (26.0%) Dysphoria high SoC *N* = 3 (3.0%) Depression low SoC *N* = 7 (8.0%) Depression high SoC *N* = 0 (0%)	Correlation between SoC and BDI: ID: $r = -0.72$, $P < .01$ TD: $r = -0.66$, $P < .01$ When adjusted for SoC, fewer ID mothers with low SoC had normal BDI scores compared with TD mothers: $F_{2,216} = 9.0$, $P < .01$	1. BDI 2. SoC	1. BDI: 4-point scale of 0–4 with score 10–20 for dysphoria and >20 for depression 2. SoC: 7-point scale, 13 items, e.g. feeling of not being in control, of no meaning to things, of disappointment, higher score indicates higher sense of coherence
2002	Weiss	*M* = 15.5, SD = 2.81	*M* = 9.2, SD = 2.16	$F = 15.95$, $P < .001$ ID mothers were more depressed than TD mothers $r = 0.27$	BDI	BDI: symptoms of depression (e.g. hopelessness, loss of interest in activities, changes in diet and sleep)

Table 3. *(Continued)*

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
2003	Emerson	Depressed to a great extent 14.0%; to some extent 30.0%	Depressed to a great extent 2.0%; to some extent 7.0%	Mann–Whitney $z = 17.93$, $P < .001$	SDQ	Self-assessed psychological impact of the difficulties of the sampled child/ adolescent on the respondent
2005	Eisenhower *et al.*	$M = 10.4$, SD = 7.6	$M = 9.6$, SD = 9.0	Not reported	CES-D	CES-D questionnaire to measure depressive symptoms in adults; higher scores indicate higher level of depressive symptoms
2006	Abasiubong *et al.*	$N = 11$ (10.4%) with score ≥8	$N = 4$ (3.9%) with score ≥8	not reported	HADS	HADS: depression score ≥8
2006	Olsson & Hwang	$M = 9.7$, SD = 7.9 Depression in relation to participation in work: <20 h/ week $M = 11.4$, SD = 10.0 20–33 h/ week $M = 8.8$, SD = 6.5 >33 h/week $M = 8.2$, SD = 6.7	$M = 5.3$, SD = 6.1 Depression in relation to participation in work: <20 h/week $M = 7.8$, SD = 7.5 20–33 h/week $M = 5.0$, SD = 5.8 > 33 h/week $M = 4.3$, SD = 5.2	Not reported. ID mothers more often work <20 hours a week than TD mothers χ^2 (2, $n = 352$) = 25.5, $P < .05$	BDI	BDI: 4-point scale of 0–4 with score 10–20 for dysphoria and >20 for depression
2006	Emerson *et al.*	Happy: not at all 1.2%; not very 8.3%	Happy: not at all 0.5%; not very 3.3%	Mothers of children with ID significantly less happy: Mann–Whitney $z = 4.71$, $P < .001$. Association between caring for a child with ID and maternal happiness: OR = 1.42, $P < .01$. Impact of controlling for maternal characteristics on the association between caring for a child with ID and maternal happiness: OR = 1.00, *P* not reported. Maternal general health not good: OR = 2.24, $P < .001$	4-point scale on a self-completion questionnaire	Self-report of feeling unhappy
2008	Olsson & Hwang	1. $M = -12.4$, SD = 13.9 High wellbeing $N = 42$ (62.7%) Medium wellbeing $N = 13$ (21.0%)	1. $M = -20.9$, SD = 12.9 High wellbeing $N = 153$ (86.0%) Medium wellbeing $N = 23$ (12.9%)	1. More ID mothers scored above the cut-off point for low wellbeing λ (2) = 16.5, $P = .01$ $F_{1,429} = 13.5$, $P = .03$	1. BDI-2r 2. Self-rated health scale	1. BDI-2r: 7-point scale of −3 (*could not be happier*) to 3 (*could not be more unhappy*) High wellbeing (BDI −6|−9) Medium wellbeing (BDI −8–8) Low wellbeing (BDI 9–61)

Table 3. *(Continued)*

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
		Low wellbeing N = 7 (11.3%) 2. Poor self-rated general health: *N* = 25 (40.3%)	Low wellbeing *N* = 2 (1.1%) 2. Poor self-rated general health: *N* = 26 (14.6%)	2. Strongest predictor of depressive symptoms was overall self-rated health; higher scores on health correlated strongly with high self-reported wellbeing on BDI $\beta = .37$, $P < .001$; for mothers ID only $\beta = .27$, $P < .01$		2. 4-point health scale from excellent to poor
2011	Mirsaleh *et al.*	*M* = 4.52, SD = 4.98	*M* = 1.43, SD = 1.98	$t = 6.42$, $P < .001$	GHQ-28	GHQ-28: symptoms of severe depression; higher score indicates worse health
2013	Kilic *et al.*	Special education X = 15.92, SS = 5.8 Mainstream education X = 17.61, SS = 6.6	X = 14.77, SS = 6.9	$F = 3.521$, $P = .031$	BDI	BDI: not reported
2013	Norlin & Broberg	T1: *M* = −12.4, SD = 13.8 T2: *M* = −14.5, SD = 13.0	T1: *M* = −20.8, SD = 12.9 T2: *M* = −20.9, SD = 14.3	ID vs. TD mothers: T1: $F = 10.18$, $P < .001$ T2: not reported for mothers separately. Child ID status contributed significantly to wellbeing in mothers $t = 3.18$, $P < .01$	BDI-2r	BDI-2r: 7-point scale of −3 (*could not be happier*) to 3 (*could not be more unhappy*); range −63 to 63 with higher scores for poorer wellbeing
2015	Long *et al.*	Not reported	Not reported	Significant group differences with highest levels of depressive symptoms observed in Latina ID mothers: $F_{3,188} = 4.92$, $P = .003$	BSI: depressive symptoms scale	5-point scale
2018	AlAnsari and Jahrami	*M* = 1.46, SD = 0.3	*M* = 0.86, SD = 0.19	$P = .270$	GHQ-28	GHQ-28: symptoms of severe depression; higher score indicates worse health
Studies of mothers of children and adults						
2015	Fairthorne *et al.*	Affective disorders: mild–moderate ID: 306 (6.6%); severe ID: 14 (4.4%)	Affective disorders: 7881 (2.9%)	Mild–moderate ID: IRR = 1.98, 95% CI [1.4, 2.8]; severe ID: IRR = 5.12, 95% CI [1.4, 18.5]	ICD-9 and ICD-10 codes	Statutory state-based registries of hospital admissions and outpatient mental health appointmen

Abbreviations: BDI, Beck Depression Inventory; BSI, Brief Symptom Inventory; CES-D, Centre for Epidemiological Studies Depression Scale; DIGS, Diagnostic Interview for Genetic Studies; GHQ, General Health Questionnaire; HADS, Hospital Anxiety and Depression Scale; ID, Children with intellectual disability; *M*, mean score; MMPI, Minnesota Multiphasic Personality Inventory; PSI, Parenting Stress Index; SD, standard deviation; SDQ, Strengths and Difficulties Questionnaire; SMI, serious mental illness; SoC, Sense of Coherence Scale; SS, sum of squared deviations; TD, typically developing children.

1 only. For two papers that included more than one group of mothers of children with IDs (Olsson and Hwang 2002; Eisenhower *et al.* 2005), but only one comparison group for the typically developing population, we calculated a combined mean and standard deviation for these subgroups to minimise bias. For Olsson and Hwang (2006), only the results for the total control and comparison group were entered in the meta-analysis. The remaining results were presented by three categories of work hours and were not directly related to the study's hypothesis testing if a presence of a child with ID in the family is related to parental wellbeing. The pooled SMD for differences in depression scores between mothers of children with IDs and mothers of typically developing children was 0.77, 95% CI [0.52, 1.02], $P < .001$, showing that mothers of children with IDs have higher depression than mothers of typically developing children. However, there was a high level of heterogeneity between studies, with $I^2 = 86\%$. Due to substantial heterogeneity, a further meta-analysis was then conducted with the six eligible papers that had used the Beck Depression Inventory (Harris and McHale 1989; Norlin and Broberg 2013; Olsson and Hwang 2002, 2006, 2008; Weiss 2002), with similar results: effect size was 0.84 95% CI [0.50, 1.18], $P < .001$, with high heterogeneity: $I^2 = 87\%$.

Other indicators of mental ill-health

Twenty-three studies investigated other indicators of mental ill-health (Table 4); 20 on mothers of children, two on mothers of children and adults, and one on mothers of adults. Five used the General Health Questionnaire (Emerson 2003; Abasiubong *et al.* 2006; Mirsaleh *et al.* 2011; Totsika *et al.* 2011b; AlAnsari and Jahrami 2018), four used the Family Impact Questionnaire (Eisenhower *et al.* 2005; Neece and Baker 2008; Norlin and Broberg 2013; Blacher and Baker 2019) and two used records in statutory state-based registries of hospital admissions and outpatient mental health appointments (Fairthorne *et al.* 2015, 2016). Fifteen studies used a variety of other instruments, sometimes in conjunction with those already mentioned. The studies have been divided into the categories of stress, emotional burden, common mental disorders, somatic symptoms, psychiatric disorders and wellbeing.

Stress

Stress was investigated in eight studies on mothers of children (Ong *et al.* 1999; Eisenhower *et al.* 2005; Neece and Baker 2008; Gupta and Kaur 2010; Totsika *et al.* 2011a; Norlin and Broberg 2013; Hosseininik *et al.* 2018; Blacher and Baker 2019). Four used the Family Impact Questionnaire (Eisenhower *et al.* 2005; Neece and Baker 2008; Norlin and Broberg 2013; Blacher and Baker 2019). The remaining studies used a variety of measures.

Data were suitable for meta-analysis in six studies, which measured stress (Ong *et al.* 1999; Eisenhower *et al.* 2005; Gupta and Kaur 2010; Norlin and Broberg 2013; Hosseininik *et al.* 2018; Blacher and

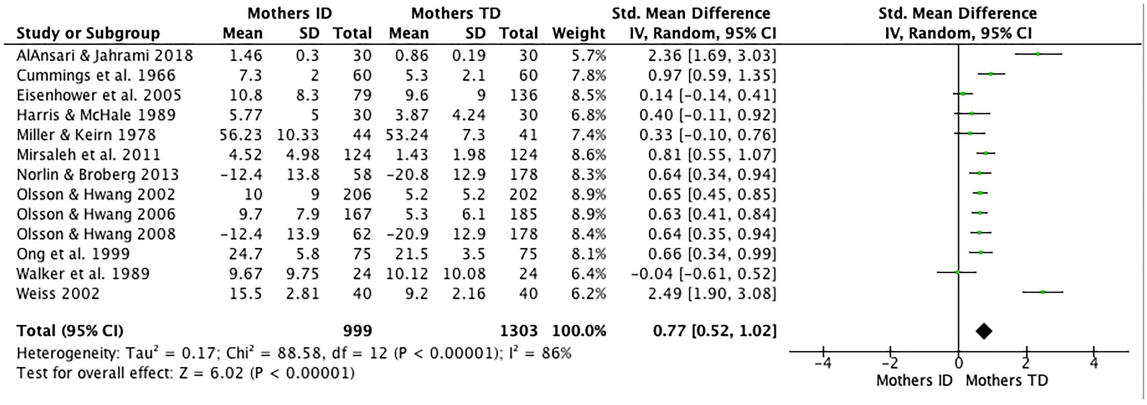

Figure 3. Forest plot for 13 analysable studies presenting findings on depression in mothers of people with intellectual disabilities. Shows standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]

Table 4 Studies investigating other indicators of mental ill-health in mothers of children with intellectual disabilities

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
Stress						
Studies of mothers of children						
1999	Ong *et al.*	Total PSI: $M = 274$; SD = 42.5 Child domain; $M = 132.9$, SD = 23.6 Parent domain: $M = 141.4$, SD = 26.1 Social isolation: $M = 14.1$, SD = 3.9 Health problems $M = 12.4$, SD = 3.3 Life stress: $M = 8.7$, SD = 7.9	Total PSI: $M = 232.1$, SD = 31.8 Child domain: $M = 106.8$, SD = 15.7 Parent domain $M = 126.5$, SD = 16.8 Social isolation: $M = 13.5$, SD = 2.4 Health problems: $M = 11.5$, SD = 2.4 Life stress: $M = 7.5$, SD = 7.2	Total PSI: $P < .001$ Child domain: $P < .001$ Parent domain: $P < .001$ Social isolation: $P = .271$ Health problems: $P = .058$ Life stress: $P = .338$	PSI	Child domain (CDS) and parent-domain stress (PDS), life stress and a total PSI score
2005	Eisenhower *et al.*	FIQ negative impact $M = 18.3$, SD = 11.5	FIQ negative impact $M = 11.1$, SD = 8.0	$B = -1.47$, SE = 2.52, $\beta = -0.06$	FIQ	FIQ social relationships and negative feelings about parenting subscale combined score with higher scores indicating higher level of stress
2008	Neece & Baker	Not reported	Not reported	Negative impact subscale: ID mothers reported more parenting stress at child's age 6: ($t = 4.41$, $P < .001$) and at child's age 8 ($t = 7.06$, $P < .001$)	FIQ	FIQ: 4-point scale from not at all to very much; higher score indicates higher level of stress
2010	Gupta & Kaur	Mental stress > physical stress in 21/30 mothers; highest mental stress score for all parents was 32; total stress score for all ID parents $M = 32.16$, SD = 10.80	Mental stress > physical stress in 7/13 mothers; highest mental stress score for all parents was 26; total stress score for all TD parents $M = 20.50$, SD = 5.00	Total stress score: $t = 2.46$; $P < .01$	QSAT	Maximum possible score for mental health is 38; stressor occurs more than *once a week* = 2, *every month* = 1, *less frequently than once a month* = 0
2011a	Totsika *et al.*	1. SMI: 9.26%, presence of ID OR = 1.72, 95% CI [1.14–2.59] 2. Maternal psych. distress: 12.56%, presence of ID OR = 1.11, 95% CI [0.83–1.48]	1. SMI: 3.18% 2. Maternal psych. distress: 8.09%	1. SMI: ID vs. TD $F = 32.57$, $P < .001$ 2. Maternal psych. distress: ID vs. TD $F = 10.47$, $P < .001$	1. K6 2. SF-8 Mental Component Summary	1. K6: self-report of psychological distress on a scale 0–4 with cut-off of ≥13 for SMI 2. SF-8: mental health functioning on 1–6 scale, lower scores indicate frequent psychological distress and higher scores indicate frequent positive affect

Table 4. *(Continued)*

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
2013	Norlin & Broberg	Parenting stress: *M* = 10.8, SD = 8.2	Parenting stress: *M* = 4.6, SD = 3.8	Group differences not reported for mothers separately	FIQ (16/50 items)	FIQ: 4-point scale from not at all to very much; higher score indicates higher level of stress
2018	Hosseininik *et al.*	Tolerance *M* = 7, SD = 1.16 Absorption *M* = 5, SD = 1.16 Evaluation *M* = 9.33, SD = 0.95 Adaptation *M* = 9.50, SD = 0.96	Tolerance *M* = 7.50, SD = 0.96 Absorption *M* = 5.33, SD = 0.95 Evaluation *M* = 9.83, SD = 0.90 Adaptation *M* = 10, SD = 0.82	Difference in at least one of the components: $F_{4,91} = 41.91$, $P = .001$ Tolerance: $F_{1,94} = 5.22$, $P = .025$ Absorption: $F_{1,94} = 2.35$, $P = .12$ Evaluation: $F_{1,94} = 6.94$, $P = .01$ Adaptation: $F_{1,94} = 7.42$, $P = .008$	DTS	Emotional distress tolerance self-evaluation index with 4 subscales
2019	Blacher & Baker	*M* = 21.6, SD = 14.2	*M* = 10.0, SD = 8.4	$F = 29.94$, $P < .001$	FIQ	FIQ: 4-point scale from not at all to very much; higher score indicates higher level of stress
Emotional burden						
Studies of mothers of children						
1992	Walker *et al.*	Personal burden: *M* = 3.62, SD = 1.28 Life-span care: *M* = 4.79, SD = 1.18 Lack of personal reward: *M* = 0.42, SD = 0.58 Terminal illness stress: *M* = 0.75, SD = 0.99 Preference for institutional care: *M* = 0.71, SD = 0.69	Personal burden: *M* = 2.83, SD = 1.27; remaining scales not administered to mothers of TD children because they refer to care of a family member with a chronic condition	Significant group differences found for: life-span care $F_{2, 62} = 21.94$, $P < .001$; terminal illness stress $F_{2, 62} = 30.99$, $P < .001$; Total score $F_{2, 62} = 4.72$, $P < .01$. Correlation of ID with: personal burden $r = .15$, P not reported; life-span care $r = .35$, $P < .001$; lack of personal reward $r = -.12$, P not reported; terminal illness stress $r = -.22$, $P < .05$; preference for institutional care $r = -.25$, $P < .05$. Correlations of depression with: personal burden $r = .50$, $P < .001$; life-span care $r = .16$, P not reported; lack of personal reward $r = -.07$, P not reported; terminal illness stress $r = .30$, $P < .01$; preference for institutional care $r = -.20$, $P < .05$; ID $r = -.16$, P not reported	QRS-S	QRS-S 5 scales on respondent attitudes with 0–6 score range per scale

Table 4. *(Continued)*

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
2002	Weiss	Emotional exhaustion: M = 27.75, SD = 2.42	Emotional exhaustion: M = 23.75, SD = 2.95	Emotional exhaustion: F = 3.83, P = .024	MBI	Emotion emotional exhaustion subscale
2003	Emerson	Tiredness to a great extent 24.0%; to some extent 31.0%	Tiredness to a great extent 4.0%; to some extent 8.0%	Mann–Whitney z = 20.17, $P < .001$	SDQ	Self-assessed psychological impact of the difficulties of the sampled child/adolescent on the respondent
2005	Lenhard *et al.*	Not reported	Not reported	Emotional strain: significantly elevated score in ID mothers compared with the TD mothers (P = .010)	Questionnaire from an unpublished preliminary study	6-point-scale ranging from 1 (*totally disagree*) to 6 (*totally agree*); subscale scores were computed with values ranging between 0 and 1
Common mental disorders						
Studies of mothers of children						
2003	Emerson	M = 2.6; 35% with GHQ > 2	M = 1.8; 25% with GHQ score >2	Mann–Whitney z = 13.9, df = 1; $P < .001$	GHQ-12	GHQ > 2
2006	Abasiubong *et al.*	N = 28 (26.4%) with GHQ score ≥5	N = 10 (9.9%) with GHQ score ≥5	Not reported	GHQ-28	GHQ-28: score ≥5
2007	Mugno *et al.*	Psychological subdomain: M = 68.79, SD = 13.40	Psychological subdomain: M = 64.38, SD = 15.40	Psychological subdomain: F = 4.17, P = .007	WHOQOL-BREF	Psychological subdomain: self-perception of mental state and wellbeing: higher score indicates better quality of life in psychological domain
2011	Mirsaleh *et al.*	Total score: M = 27.29, SD = 14.73	Total score: M = 17.49, SD = 7.07	Total score: t = 6.68, $P < .001$	GHQ-28	GHQ-28: total score; higher score indicates worse health
2011b	Totsika *et al.*	Maternal emotional disorder: 32.4%	Maternal emotional disorder: 23.7%	ID presence compared with TD OR = 1.02, 95% CI [.84–1.24]; χ^2 = 40.12, $P < .001$ V = 0.047	GHQ-12	GHQ-12: screening for common mental disorders
2018	AlAnsari and Jahrami	Total score: M = 6.8, SD = 1.1	Total score: M = 4.6, SD = 0.90	Not reported	GHQ-28	GHQ-28: total score; higher score indicates worse health
2019	Blacher & Baker	M = 25.9, SD = 24.5	M = 14.2, SD = 14.3	F = 13.19, $P < .001$	SCL	SCL: 4-point scale from not at all to extremely; higher score indicates higher level of adult mental health symptoms (somatisation, interpersonal sensitivity, anxiety, depression, and hostility); total score range is 0–140

Table 4. *(Continued)*

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
Somatic symptoms						
Studies of mothers of children						
2002	Weiss	Somatic complaints: *M* = 15.09, SD = 3.15	Somatic complaints: *M* = 13.41, SD = 2.55	Somatic complaints: $F = 13.06$, $P < .001$	Questionnaire developed by the US Department of Health, Education and Welfare	10 questions on stress-related symptoms (e.g. sweaty hands, tachycardia and shortness of breath)
2011	Mirsaleh *et al.*	Somatic symptoms: *M* = 7.06, SD = 4.08	Somatic symptoms: *M* = 4.40, SD = 2.95	Somatic symptoms: $t = 5.89$, $P < .001$	GHQ-28	GHQ-28: somatic symptoms subscale; higher score indicates worse health
2015	Long *et al.*	Not reported	Not reported	Significant group differences with highest levels of GSI and somatic symptoms observed in Latina ID mothers: GSI $F_{3,188} = 3.30$, $P = .02$; somatic symptoms $F_{3,188} = 3.98$, $P = .009$	BSI: Global Severity Index (GSI) and somatic symptoms scales	5-point scale
2018	AlAnsari and Jahrami	Somatic symptom: *M* = 1.13, SD = 0.3	Somatic symptom: *M* = 1.46, SD = 0.39	Somatic symptom: $P = .501$	GHQ-28	GHQ-28: somatic symptoms subscale; higher score indicates worse health
Psychiatric disorders						
Studies of mothers of children						
1978	Miller & Keirn	Hypochondriasis: *M* = 52.75, SD = 10.37 Hysteria: *M* = 59.25, SD = 10.95 Psychopathic deviate: *M* = 58.91, SD = 11.16 Paranoia: *M* = 54.90, SD = 8.97 Schizophrenia: *M* = 55.73, SD = 8.89 Hypomania: *M* = 55.75, SD = 9.50	Hypochondriasis: *M* = 53.40, SD = 9.73 Hysteria: *M* = 58.19, SD = 7.59 Psychopathic deviate: *M* = 56.84, SD = 7.15 Paranoia: *M* = 55.05, SD = 8.54 Schizophrenia: *M* = 34.76, SD = 7.05 Hypomania: *M* = 50.08, SD = 7.94	Hypomania: $t = 2.93$, $P < .005$; *t*-test results not reported for other scales	MMPI	Elevations reflect parents' pathological reactions to the presence of a disabled child; higher score indicates worse mental health

Table 4. *(Continued)*

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
Studies of mothers of children and adults						
2015	Fairthorne *et al.*	Any psychiatric disorder: Mild–moderate ID: 890 (19.2%) Severe ID: 52 (16.2%)	Any psychiatric disorder: 25 818 (9.5%)	Mild–moderate ID: IRR = 1.80, 95% CI [1.5, 2.2] Severe ID: IRR = 0.85, 95% CI [0.4, 1.9]	ICD-9 and ICD-10 codes	Statutory state-based registries of hospital admissions and outpatient mental health appointments
2016	Fairthorne *et al.*	Mothers of children with ID spent 7% more days in hospital compared with mothers of TD children	-	Any psychiatric disorder: IRR = 1.73 (1.4, 2.1) Inpatient psychiatric episode treatment: IRR = 1.07 (1.05, 1.10)	ICD-9 and ICD-10 codes	Statutory state-based registries of hospital admissions and outpatient mental health appointments
Wellbeing						
Studies of mothers of children						
2002	Olsson & Hwang	Sense of coherence: ID M = 64.4, SD = 14.5	Sense of coherence: M = 69.1, SD = 11.9	Mothers ID had lower SoC than mothers TD $F_{2,408}$ = 11.3, $P < .01$	SoC	SoC: 7-point scale, 13 items, e.g. feeling of not being in control, of no meaning to things, of disappointment, higher score indicates higher sense of coherence
2007	Mugno *et al.*	Overall quality of life: M = 63.27, SD = 20.48	Overall quality of life: M = 77.98, SD = 13.73	F = 9.16, $P < .001$	WHOQOL-BREF	Overall perceived quality of life; higher score indicates better quality of life
2011	Mirsaleh *et al.*	Social dysfunction: M = 7.68, SD = 3.63	Social dysfunction: M = 6.10, SD = 2.64	Social dysfunction: t = 3.92, $P < .001$	GHQ-28	GHQ-28: social dysfunction subscale; higher score indicates worse health
2018	AlAnsari and Jahrami	1. Social dysfunction: M = 1.46, SD = 0.2 2. Psychological health: M = 70.3, SD = 2.6 3. GAF mean score: M = 72.61, SD = 2.9	1. Social dysfunction: M = 1.06, SD = 0.28 2. Psychological health: M = 72.9, SD = 2.2 3. GAF mean score: M = 88.73, SD = 0.60	1. Social dysfunction: P = .392 2. Psychological health: P = .024 3. P = .001	1. GHQ-28 2. WHOQOL-BREF 3. GAF	1. Social dysfunction domain 2. Psychological health domain 3. Scores range of 0–100: 71–80 = symptoms are present, slight impairment in social, occupational, or school functioning; 81–90 = absent or minimal symptoms, good functioning in all areas
2018	Hosseininik *et al.*	Emotional adjustment: M = 9.50, SD = 0.96 Health adjustment: M = 6.16, SD = 0.69	Emotional adjustment: M = 10.83, SD = 1.07 Health adjustment: M = 6.66, SD = 0.75	Difference in at least one of the components: $F_{5,90}$ = 23.64, $P < .001$) Emotional adjustment: $F_{1,94}$ = 40.64, P = .001 Health adjustment: $F_{1,94}$ = 11.43, P = .001	Bell Adjustment Inventory	5 components of job matching, home-based, emotional, health and social adjustment; 32 questions with answers of good/I do not know

Table 4. *(Continued)*

Year	Author	Mothers ID	Mothers TD	Statistical comparison	Measures used	Definition of mental ill-health
Studies of mothers of adults						
2001	Rimmerman & Muraver	1. Wellbeing M = 14.71, SD = 1.86 2. Poor health M = 25.80, SD = 25.82	1. Wellbeing M = 14.41, SD = 2.32 2. Poor health M = 17.90, SD = 20.51	1. No significant group differences 2. t = 2.88, df = 284, $P < .01$	1. Affect Balance Scale 2. Health Scale of the Undesired Daily Life Events	1. 5-point scale on Affect Balance Scale, higher score indicates better wellbeing 2. Self-report of episodic or chronic states; higher score indicates poorer health

Abbreviations: BDI, Beck Depression Inventory; BSI, Brief Symptom Inventory; DIGS, Diagnostic Interview for Genetic Studies; DTS, Distress Tolerance Questionnaire; FIQ, Family Impact Questionnaire; GAF, Global Assessment of Function; GHQ, General Health Questionnaire; ID, children with intellectual disability; IRR, incidence risk ratio; *M*, mean score; MBI, Maslach Burnout Inventory; MMPI, Minnesota Multiphasic Personality Inventory; PSI, Parenting Stress Index; PSQ, Perceived Stress Questionnaire; QRS-S, Questionnaire on Resources and Stress-Short Form; QSAT, Quick Stress Assessment; SCL, Hopkins Symptoms Checklist; SD, standard deviation; SDQ, Strengths and Difficulties Questionnaire; SE, standard error for the unstandardised beta; SMI, Serious Mental Illness; SoC, Sense of Coherence Scale; TD, typically developing children; WHOQOL-BREF, World Health Organization Quality of Life Short Version Instrument.

Baker 2019) (Fig. 4). Norlin and Broberg (2013) collected data at two time points, but data on stress were available only for time 1. Hosseininik *et al.* (2018) collected data on four different subscales of the Distress Tolerance Questionnaire, but only results for the tolerance to stress subscale were entered in the meta-analysis. For Ong *et al.* (1999), only a total score on the Parenting Stress Index was entered in the meta-analysis. The pooled SMD for differences in stress levels between mothers of children with IDs and mothers of typically developing children was 0.82, 95% CI [0.31, 1.33], P = .002. There was evidence of a high level of heterogeneity between studies, with I^2 = 90%, but the effect size was large, showing that mothers of children with IDs are indeed more stressed than mothers of typically developing children.

Emotional burden

Emotional burden was investigated in four studies on mothers of children (Walker *et al.* 1992; Weiss 2002; Emerson 2003; Lenhard *et al.* 2005), using a variety of measures, including an unvalidated questionnaire from an unpublished preliminary study (Lenhard *et al.* 2005).

Two of the papers reported data suitable for meta-analysis on emotional burden/exhaustion (Walker *et al.* 1992; Weiss 2002). The pooled SMD for differences in levels of emotional burden/exhaustion in mothers of children with IDs and mothers of typically developing children was 1.05, 95% CI [0.21, 1.89], P = .01, showing a statistically significant large effect size demonstrating that levels of emotional burden/exhaustion were higher in mothers of children with IDs compared with mothers of other children. However, there was evidence of high statistical heterogeneity between studies, with I^2 = 79% (Fig. 5).

Common mental disorders

Common mental disorders were investigated in seven studies on mothers of children (Emerson 2003; Abasiubong *et al.* 2006; Mugno *et al.* 2007; Mirsaleh *et al.* 2011; Totsika *et al.* 2011b; AlAnsari and Jahrami 2018; Blacher and Baker 2019). Five used the General Health Questionnaire (Emerson 2003; Abasiubong *et al.* 2006; Mirsaleh *et al.* 2011; Totsika *et al.* 2011b; AlAnsari and Jahrami 2018). All reported

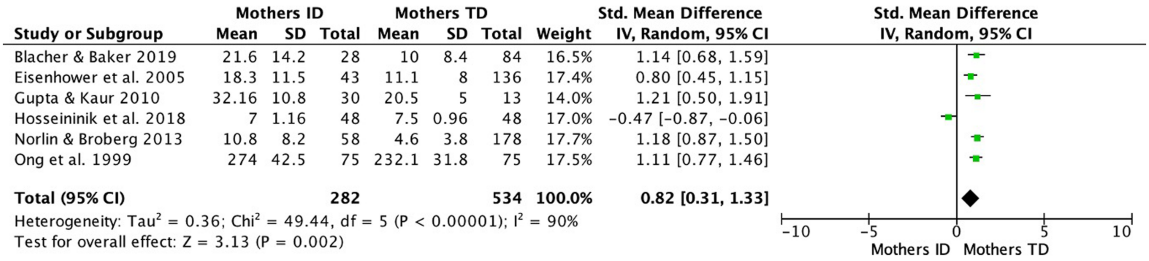

Figure 4. Forest plot for six analysable studies presenting findings on stress in mothers of people with intellectual disabilities. Shows standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]

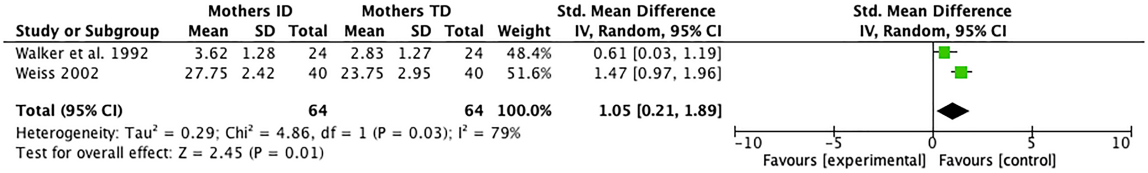

Figure 5. Forest plot for two analysable studies presenting findings on emotional burden/exhaustion in mothers of people with intellectual disabilities. Shows standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]

higher level of mental disorders in mothers of people with IDs.

Data were suitable for meta-analysis in four studies, which examined common mental disorders (Mugno *et al.* 2007; Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018; Blacher and Baker 2019) (Fig. 6). The pooled SMD for difference in occurrence of common mental disorders in mothers was 0.94, 95% CI [0.37, 1.52], $P < .001$, showing a large effect size demonstrating that levels of common mental disorders were higher in mothers of children with IDs compared with mothers of typically developing children. However, there was evidence of high statistical heterogeneity between studies, with $I^2 = 87\%$.

Somatic symptoms

Somatic symptoms were investigated in four studies on mothers of children (Weiss 2002; Mirsaleh *et al.* 2011; Long *et al.* 2015; AlAnsari and Jahrami 2018). Two studies used the somatic symptoms subscale of the General Health Questionnaire (Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018) while the remaining studies used a variety of measures. All studies reported more somatic symptoms for mothers of people with IDs apart from one paper (AlAnsari and Jahrami 2018).

Data were suitable for meta-analysis in three studies which investigated somatic symptoms of mental ill-health (Weiss 2002; Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018). The pooled SMD for level of somatic symptoms experienced due to mental ill-health by mothers of children with IDs and mothers of typically developing children was 0.15, 95% CI [−0.77, 1.07], $P > .1$, showing a statistically non-significant small effect size demonstrating that levels of these somatic symptoms do not differ greatly between mothers of children with IDs and mothers of typically developing children. There was also evidence of high statistical heterogeneity between studies, with $I^2 = 94\%$ (Fig. 7).

Psychiatric disorders

Any psychiatric disorder was investigated in three studies; one on mothers of children (Miller and Keirn 1978) and two on mothers of children and adults (Fairthorne *et al.* 2015, 2016). Two studies used records in statutory state-based registries of hospital admissions and outpatient mental health appointments (Fairthorne *et al.* 2015, 2016). All three

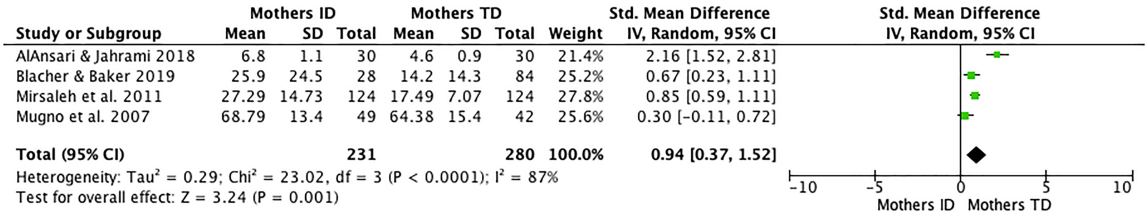

Figure 6. Forest plot for four analysable studies presenting findings on common mental disorders in mothers of people with intellectual disabilities. Shows standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]

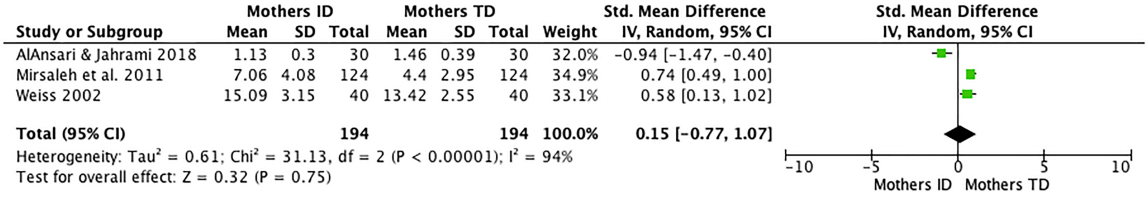

Figure 7. Forest plot for three analysable studies presenting findings on somatic symptoms in mothers of people with intellectual disabilities. Shows Standardised mean difference (SMD) and 95% confidence interval (CI). Pooled effects estimate shown for random-effects model. ID, children with intellectual disability; TD, typically developing children; SD, standard deviation. [Colour figure can be viewed at wileyonlinelibrary.com]

studies reported higher levels of psychiatric disorders in mothers of people with IDs, with the exception of hypochondriasis and paranoia subscales investigated by Miller and Keirn (1978), which reported lower rates. Only one study reported data suitable for meta-analysis, so it was not possible to conduct one.

Wellbeing

Six studies investigated wellbeing, using different measures; five on mothers of children (Olsson and Hwang 2002; Mugno *et al.* 2007; Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018; Hosseininik *et al.* 2018) and one on mothers of adults (Rimmerman and Muraver 2001). Two studies used the General Health Questionnaire to measure social dysfunction (Mirsaleh *et al.* 2011; AlAnsari and Jahrami 2018) while the remaining studies used a variety of measures to investigate different concepts of wellbeing. One study reported non-significant better wellbeing ratings in mothers of children with IDs compared with mothers of typically developing children (Mugno *et al.* 2007). Another study found no statistical difference in wellbeing for mothers of adults with IDs compared with typically developing adults (Rimmerman and Muraver 2001). Although all studies reported data suitable for meta-analysis, they investigated very diverse and not easily comparable concepts of wellbeing such as sense of coherence, overall quality of life, social dysfunction, psychological health, emotional and health adjustment, wellbeing, and poor health. For this reason, data on wellbeing were not meta-analysed.

Gaps in evidence base

Only three studies, which met our inclusion criteria, investigated mental ill-health of mothers of adults with IDs, including two by the same author utilising the same statutory state-based registries of hospital admissions and outpatient mental health appointments, which displayed a low risk of bias (Fairthorne *et al.* 2015, 2016). The third study, which investigated mental ill-health in mothers of adults only, displayed an uncertain risk of bias, including a small and non-representative sample and a report of no statistical differences observed between the studied populations, thus limiting its conclusions (Rimmerman and Muraver 2001). These studies provide much needed insight into the comparative health of mothers of older people with

IDs, but clearly, more evidence is needed, particularly given the likely increased burden of care arising with ageing-related health difficulties in both mothers and their adult children (Haley and Perkins 2004; Chou *et al.* 2011). As for all the studies reviewed, findings were not broken down by age group of mothers or their children or stages in the caregiving trajectory.

Discussion

This systematic review extends the previous narrative review of studies without comparison groups (O'Keeffe and O'Hara 2008) by providing direct meta-analytic evidence that mental ill-health is more common in mothers of people with IDs than in mothers of typically developing people. This includes higher rates of anxiety, depression, parenting stress, common mental disorders and emotional burden, but not somatic symptoms. However, we must acknowledge the heterogeneity of the included studies, impeding syntheses in this systematic review and in turn, increasing the need for further robust research in this area. Our findings are consistent with previous reports on mothers of children with various disabilities or chronic health conditions (Singer 2006; Bailey *et al.* 2007; Miodrag *et al.* 2015) and developmental disabilities (Lee 2013). The ill-health experienced by mothers of people with IDs is highly likely to also impact their children and create a circular vortex of family ill-health, as studies of samples with different disabilities (Witt *et al.* 2003) and in general population (Plass-Christl *et al.* 2017) have shown. We also identified considerable gaps in the evidence base, particularly a lack of robust, comparative studies on mothers' mental ill-health across the caregiving trajectory, which is important given that typically developing children usually become increasingly independent and move on to a much greater extent than children with IDs. Information on mothers' health is important to identify the pivotal points in the caregiving trajectory when mothers require support to prevent/ameliorate mental ill-health, and to heighten awareness of burden of care for healthcare providers to ensure that families of people with IDs receive the best possible care.

Current evidence and future directions

Alhough we identified a number of studies, the quality of existing research is limited, particularly regarding sample sizes, sample recruitment, and inclusion criteria. Unlike in a meta-analysis of studies investigating adverse health in parents of children with disabilities and chronic health conditions using the Parenting Stress Index's Health Sub-domain only (Miodrag *et al.* 2015), a wide variety of mental ill-health measures and concepts were used in this meta-analysis, to a certain extent impeding synthesis. A meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities also recorded a variety of measures used to study the concept, but this fact did not impede the pooling of the effect sizes (Singer 2006).

As noted above, few studies that met our inclusion criteria investigated mental ill-health in mothers of adults, and none reported findings separately for mothers of different ages or at different stages of the caregiving trajectory. More robust evidence is, therefore, needed in order to further explore and isolate with certainty the primary causes of parental ill-health in order to draw firm conclusions and inform interventions, as well as improving our understanding of the long-term impact of the mental ill-health experienced across the caregiving trajectory on quality of life, suicidal ideation and other health inequalities experienced by mothers of people with IDs. This in turn will allow for informed decision making around support and healthcare service commissioning and provision for families of people with IDs. Given that maternal mental health difficulties are likely to have a substantial impact on family needs and quality of life, both the need for improved support for mothers and the need for further methodologically robust research is clearly indicated.

It is also worth noting that while our review did not focus on the factors, which may have particular impact on mothers' mental health, these may include a multitude of components such as increased demands on parents' time because of the increased needs to navigate services and advocate for child's care (e.g. Haveman *et al.* 1997), awareness of and experience of stigma (e.g. Ali *et al.* 2012) or cognition of parenting self-esteem and parental locus of control

(e.g. Hill and Rose 2009). Some studies have also shown that behaviour problems in populations with IDs may contribute more significantly to parenting stress than the diagnosis of IDs per se (e.g. Baker *et al.* 2003; Neece and Baker 2008). The aforementioned studies highlight that although the findings consistently show more mental health problems in mothers of people with IDs, the aetiology of these problems may not be solely attributed to the diagnosis of IDs itself but is most likely associated with wider challenges of raising a child with IDs.

Strengths and limitations of this study

Strengths of our review include the prospective registration of the protocol, clear inclusion and exclusion criteria, comprehensive search strategy, including searching multiple databases and no time limits, double rating of paper selection and quality, systematic data extraction, and meta-analyses where possible. However, the review is limited by excluding papers that were not available in English. We also excluded studies with comparison groups with mothers of non-typically developing children, syndrome-specific studies, and studies on samples with developmental delay that did not clearly have IDs. Only 10% of papers were screened by two reviewers (E. R. and K. D.), but there was a perfect agreement between them (κ = 1.00) and we ensured achieving consistency prior to ceasing double rating. Data extraction was performed only by the first reviewer (E. R.) but was undertaken systematically using a structured database. We were not able to provide details on the age of the children as the majority of the reviewed studies did not report this in detail; hence, we were only able to group reviewed studies by broad developmental stages (i.e. childhood and adulthood). Our aim to determine maternal health across the caregiving trajectory was undermined by the limitations of the evidence base. Methodological, outcome measure and reporting heterogeneity limited the studies that could be included in the meta-analyses and conclusions that could be drawn from them. Indeed, even after restricting the number of synthesised studies, heterogeneity was high in all meta-analyses, and this limitation on reliability of results must be acknowledged. Furthermore, the small number of studies, which reported on the mean, standard deviation and sample size necessary for calculating an unbiased SMD meant that findings from meta-analyses of studies on anxiety, common mental disorders, somatic symptoms of mental ill-health and emotional burden/exhaustion are limited to very few studies. Additionally, included studies were predominantly from high-income countries and included samples of predominantly Caucasian mothers, so there may be unexplored cultural differences in the reviewed studies (e.g. Eisenhower and Blacher 2006; Blacher and Baker 2007; Long *et al.* 2015). For these reasons, findings from these meta-analyses should be interpreted with caution.

Conclusions

Mothers of people with IDs experience more mental ill-health than mothers of typically developing people, particularly anxiety, depression, parenting stress, common mental disorders and emotional burden. Studies are heterogeneous, and gaps remain, particularly on mothers of adults with IDs, and at different ages and stages of the caregiving trajectory. Healthcare providers require heightened awareness of the high rates of mental ill-health and burden of care placed on mothers of people with IDs and need to provide support for the whole family, not only the person with IDs.

Acknowledgements

This work was funded through the Economic and Social Research Council: ES/P010539/1.

Source of Funding

This work was funded through the Economic and Social Research Council (grant award number ES/P010539/1).

Conflict of Interest

No conflicts of interest have been declared.

Data Availability Statement

Not applicable, as this study involved a systematic review only.

References

Abasiubong F., Obembe A. & Ekpo M. (2006) A controlled study of anxiety and depression in mothers of children with learning disability in Lagos, Nigeria. *Nigerian Journal of Medicine: Journal of the National Association of Resident Doctors of Nigeria* **15**, 124–7.

AlAnsari A. M. & Jahrami H. A. (2018) Health of mothers of children with autism spectrum disorders and intellectual disability. *Bahrain Medical Bulletin* **40**, 157–61.

Ali A., Hassiotis A., Strydom A. & King M. (2012) Self stigma in people with intellectual disabilities and courtesy stigma in family carers: a systematic review. *Research in Developmental Disabilities* **33**, 2122–40.

Andersson E. (1993) Depression and anxiety in families with a mentally handicapped child. *International Journal of Rehabilitation Research* **16**, 165–9.

Azeem M. W., Dogar I. A., Shah S., Cheema M. A., Asmat A., Akbar M. *et al*. (2013) Anxiety and depression among parents of children with intellectual disability in Pakistan. *Journal of the Canadian Academy of Child and Adolescent Psychiatry* **22**, 290–5.

Bailey D. B., Jr., Golden R. N., Roberts J. & Ford A. (2007) Maternal depression and developmental disability: research critique. *Mental Retardation and Developmental Disabilities Research Reviews* **13**, 321–9.

Baker B. L., McIntyre L. L., Blacher J., Crnic K., Edelbrock C. & Low C. (2003) Pre-school children with and without developmental delay: behaviour problems and parenting stress over time. *Journal of Intellectual Disability Research* **47**, 217–30.

Blacher J. & Baker B. L. (2007) Positive impact of intellectual disability on families. *American Journal of Mental Retardation* **112**, 330–48.

Blacher J. & Baker B. L. (2019) Collateral effects of youth disruptive behavior disorders on mothers' psychological distress: adolescents with autism spectrum disorder, intellectual disability, or typical development. *Journal of Autism and Developmental Disorders* **49**, 2810–21.

Cairns D., Brown J., Tolson D. & Darbyshire C. (2014) Caring for a child with learning disabilities over a prolonged period of time: an exploratory survey on the experiences and health of older parent carers living in Scotland. *Journal of Applied Research in Intellectual Disabilities* **27**, 471–80.

Cairns D., Tolson D., Brown J. & Darbyshire C. (2012) The need for future alternatives: an investigation of the experiences and future of older parents caring for offspring with learning disabilities over a prolonged period of time. *British Journal of Learning Disabilities* **41**, 73–82.

Chen S. C., Ryan-Henry S., Heller T. & Chen E. H. (2001) Health status of mothers of adults with intellectual disability. *Journal of Intellectual Disability Research* **45**, 439–49.

Chou Y., Fu L., Lin L. & Lee Y. (2011) Predictors of subjective and objective caregiving burden in older female caregivers of adults with intellectual disabilities. *International Psychogeriatrics* **23**, 562–72.

Critical Appraisal Skills Programme (CASP) (2017) *CASP cohort study checklist*. Available at: http://www.casp-uk.net/checklists. CASP, Oxford.

Cummings S. T., Bayley H. C. & Rie H. E. (1966) Effects of the child's deficiency on the mother: a study of mothers of mentally retarded, chronically ill and neurotic children. *The American Journal of Orthopsychiatry* **36**, 595–608.

Dawson F., Shanahan S., Fitzsimons E., O'Malley G., Mac G. N. & Bramham J. (2016) The impact of caring for an adult with intellectual disability and psychiatric comorbidity on carer stress and psychological distress. *Journal of Intellectual Disability Research* **60**, 553–63.

Douma J. C. H., Dekker M. C., De Ruiter K. P., Verhulst F. C. & Koot H. M. (2006) Help-seeking process of parents for psychopathology in youth with moderate to borderline intellectual disabilities. *Journal of the American Academy of Child and Adolescent Psychiatry* **45**, 1232–42.

Eisenhower A. & Blacher J. (2006) Mothers of young adults with intellectual disability: multiple roles, ethnicity and well-being. *Journal of Intellectual Disability Research* **50**, 905–16.

Eisenhower A. S., Baker B. L. & Blacher J. (2005) Preschool children with intellectual disability: syndrome specificity, behaviour problems, and maternal well-being. *Journal of Intellectual Disability Research* **49**, 657–71.

Emerson E. (2003) Mothers of children and adolescents with intellectual disability: social and economic situation, mental health status, and the self-assessed social and psychological impact of the child's difficulties. *Journal of Intellectual Disability Research* **47**, 385–99.

Emerson E. & Hatton C. (2011) *Estimating Future Need for Social Care among Adults with Learning Disabilities in England: An Update*. Improving Health and Lives Learning Disabilities Observatory, Durham.

Emerson E., Hatton C., Llewellyn G., Blacher J. & Graham H. (2006) Socio-economic position, household composition, health status and indicators of the well-being of mothers of children with and without intellectual disabilities. *Journal of Intellectual Disability Research* **50**, 862–73.

Fairthorne J., De Klerk N. & Leonard H. (2016) Brief report: burden of care in mothers of children with autism spectrum disorder or intellectual disability. *Journal of Autism and Developmental Disorders* **46**, 1103–9.

Fairthorne J., Jacoby P., Bourke J., de Klerk N. & Leonard H. (2015) Onset of maternal psychiatric disorders after the birth of a child with intellectual disability: a retrospective cohort study. *Journal of Psychiatric Research* **61**, 223–30.

Giallo R., Seymour M., Matthews J., Gavidia-Payne S., Hudson A. & Cameron C. (2015) Risk factors associated with the mental health of fathers of children with an intellectual disability in Australia: mental health

of fathers. *Journal of Intellectual Disability Research* **59**, 193–207.

Graham F., Sinnott K. A., Snell D. L., Martin R. & Freeman C. (2013) A more "normal life": residents', family, staff, and managers' experience of active support at a residential facility for people with physical and intellectual impairments. *Journal of Intellectual and Developmental Disability* **38**, 256–64.

Gupta R. K. & Kaur H. (2010) Stress among parents of children with intellectual disability. *Asia Pacific Disability Rehabilitation Journal* **21**, 118–26.

Haley W. E. & Perkins E. A. (2004) Current status and future directions in family caregiving and aging people with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities* **1**, 24–30.

Harris V. S. & McHale S. M. (1989) Family life problems, daily caregiving activities, and the psychological well-being of mothers of mentally retarded children. *American Journal of Mental Retardation* **94**, 231.

Haveman M., van Berkum G., Reijnders R. & Heller T. (1997) Differences in service needs, time demands, and caregiving burden among parents of persons with mental retardation across the life cycle. *Family Relations* **46**, 417–25.

Higgins J. P., Thompson S. G., Deeks J. J. & Altman D. G. (2003) Measuring inconsistency in meta-analyses. *BMJ (Clinical research ed.)* **327**, 557–60.

Hill C. & Rose J. (2009) Parenting stress in mothers of adults with an intellectual disability: parental cognitions in relation to child characteristics and family support. *Journal of Intellectual Disability Research* **53**, 969–80.

Horsley S. & Oliver C. (2015) Positive impact and its relationship to well-being in parents of children with intellectual disability: a literature review. *International Journal of Developmental Disabilities* **61**, 1–19.

Hosseininik S. S., Amiri F. & Rad R. M. (2018) Comparison of distress tolerance and adjustment of mothers of intellectual disability children and mothers of normal children in Yasouj city (Iran). *NeuroQuantology* **16**, 7–13.

Innes A., McCabe L. & Watchman K. (2012) Caring for older people with an intellectual disability: a systematic review. *Maturitas* **72**, 286–95.

Jackson A. C., Liang R. P., Frydenberg E., Higgins R. O. & Murphy B. M. (2016) Parent education programmes for special health care needs children: A systematic review. *Journal of Clinical Nursing* **25**, 1528–47.

James N. (2013) The formal support experiences of family carers of people with an intellectual disability who also display challenging behaviour and/or mental health issues: What do carers say? *Journal of Intellectual Disabilities* **17**, 6–23.

Jokinen N. S. & Brown R. I. (2005) Family quality of life from the perspective of older parents. *Journal of Intellectual Disability Research* **49**, 789–93.

Kilic D., Gencdogan B., Bag B. & Arican D. (2013) Psychosocial problems and marital adjustments of families caring for a child with intellectual disability. *Sexuality and Disability* **31**, 287–96.

Lee J. (2013) Maternal stress, well-being, and impaired sleep in mothers of children with developmental disabilities: a literature review. *Research in Developmental Disabilities* **34**, 4255–73.

Lenhard W., Breitenbach E., Ebert H., Schindelhauer-Deutscher H. J. & Henn W. (2005) Psychological benefit of diagnostic certainty for mothers of children with disabilities: lessons from Down syndrome. *American Journal of Medical Genetics Part A* **133A**, 170–5.

Long K. A., Kao B., Plante W., Seifer R. & Lobato D. (2015) Cultural and child-related predictors of distress among Latina caregivers of children with intellectual disabilities. *American Journal on Intellectual and Developmental Disabilities* **120**, 145–65.

Mathie R. T., Ramparsad N., Legg L. A., Clausen J., Moss S., Davidson J. R. T. *et al*. (2017) Randomised, double-blind, placebo-controlled trials of non-individualised homeopathic treatment: systematic review and meta-analysis. *Systematic Reviews* **6**, 1–28.

Miller W. H. & Keirn W. C. (1978) Personality measurement in parents of retarded and emotionally disturbed children: a replication. *Journal of Clinical Psychology* **34**, 686–90.

Miodrag N., Burke M., Tanner-Smith E. & Hodapp R. M. (2015) Adverse health in parents of children with disabilities and chronic health conditions: a meta-analysis using the Parenting Stress Index's Health Sub-domain. *Journal of Intellectual Disability Research* **59**, 257–71.

Mirsaleh Y. R., Rezai H., Khabaz M., Afkhami Ardekani I. & Abdi K. (2011) Personality dimensions, religious tendencies and coping strategies as predictors of general health in iranian mothers of children with intellectual disability: a comparison with mothers of typically developing children. *Journal of Applied Research in Intellectual Disabilities* **24**, 573–82.

Montes G. & Halterman J. S. (2007) Psychological functioning and coping among mothers of children with autism: a population-based study. *Pediatrics* **119**, e1040–6.

Mugno D., Ruta L., D'Arrigo V. G. & Mazzone L. (2007) Impairment of quality of life in parents of children and adolescents with pervasive developmental disorder. *Health and Quality of Life Outcomes* **5**, 1–9.

Neece C. & Baker B. (2008) Predicting maternal parenting stress in middle childhood: the roles of child intellectual status, behaviour problems and social skills. *Journal of Intellectual Disability Research* **52**, 1114–28.

Norlin D. & Broberg M. (2013) Parents of children with and without intellectual disability: couple relationship and individual well-being. *Journal of Intellectual Disability Research* **57**, 552–66.

O'Keeffe N. & O'Hara J. (2008) Mental health needs of parents with intellectual disabilities. *Current Opinion in Psychiatry* **21**, 463–8.

Olsson M. B. & Hwang C. P. (2001) Depression in mothers and fathers of children with intellectual disability. *Journal of Intellectual Disability Research* **45**, 535–43.

Olsson M. B. & Hwang C. P. (2002) Sense of coherence in parents of children with different developmental disabilities. *Journal of Intellectual Disability Research* **46**, 548–59.

Olsson M. B. & Hwang C. P. (2006) Well-being, involvement in paid work and division of child-care in parents of children with intellectual disabilities in Sweden. *Journal of Intellectual Disability Research* **50**, 963–9.

Olsson M. B. & Hwang C. P. (2008) Socioeconomic and psychological variables as risk and protective factors for parental well-being in families of children with intellectual disabilities. *Journal of Intellectual Disability Research* **52**, 1102–13.

Ong L., Chandran V. & Peng R. (1999) Stress experienced by mothers of Malaysian children with mental retardation. *Journal of Paediatrics and Child Health* **35**, 358–62.

Pinquart M. & Sörensen S. (2003) Differences between caregivers and noncaregivers in psychological health and physical health: a meta-analysis. *Psychiatry and Aging* **18**, 248–56.

Plass-Christl A., Haller A., Otto C., Barkmann C., Wiegand-Grefe S., Hölling H. *et al.* (2017) Parents with mental health problems and their children in a German population-based sample: results of the BELLA study. *PLoS One* **12**, e0180410.

Rimmerman A., Gur A., Muraver M. & Yurkevich O. (2018) Personal and social life of Israeli aging mothers to adult children with intellectual disabilities: differences related to family status and living arrangement. *International Journal of Developmental Disabilities* **64**, 81–5.

Rimmerman A. & Muraver M. (2001) Experiencing undesired daily life events, instrumental functioning, social support and well-being of Israeli elderly women: comparison between caregivers/non-caregivers for adult children with mental retardation. *Journal of Women & Aging* **13**, 57–69.

Sanderson S., Tatt I. D. & Higgins J. P. T. (2007) Tools for assessing quality and susceptibility to bias in observational studies in epidemiology: a systematic review and annotated bibliography. *International Journal of Epidemiology* **36**, 666–76.

Schünemann H. J., Oxman A. D., Vist G. E., Higgins J. P. T., Deeks J. J., Glasziou P. *et al.* (2011) Chapter 12: interpreting results and drawing conclusions. In: *Cochrane Handbook for Systematic Reviews of Interventions; Version 5.1.0* (eds J. P. T. Higgins & S. Green). The Cochrane Collaboration.

Scorgie K. & Sobsey D. (2000) Transformational outcomes associated with parenting children who have disabilities. *Mental Retardation* **38**, 195–206.

Shamseer L., Moher D., Clarke M., Ghersi D., Liberati A., Petticrew M. *et al.* (2015) Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015: elaboration and explanation. *BMJ [British Medical Journal]* **349**, g7647–7.

Singer G. H. S. (2006) Meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities. *American Journal on Mental Retardation* **111**, 155–69.

Tan S. H. (2017) Assessing the needs of caregivers of children with disabilities in Penang, Malaysia. *Health & Social Care in the Community* **25**, 447–57.

Totsika V., Hastings R. P., Emerson E., Berridge D. M. & Lancaster G. A. (2011a) Behavior problems at 5 years of age and maternal mental health in autism and intellectual disability. *Journal of Abnormal Child Psychology* **39**, 1137–47.

Totsika V., Hastings R. P., Emerson E., Lancaster G. A. & Berridge D. M. (2011b) A population-based investigation of behavioural and emotional problems and maternal mental health: associations with autism spectrum disorder and intellectual disability. *Journal of Child Psychology and Psychiatry, and Allied Disciplines* **52**, 91–9.

Walker L. S., Ortiz-Valdes J. A. & Newbrough J. R. (1989) The role of maternal employment and depression in the psychological adjustment of chronically ill, mentally retarded, and well children. *Journal of Pediatric Psychology* **14**, 357–70.

Walker L. S., Van Slyke D. A. & Newbrough J. R. (1992) Family resources and stress: a comparison of families of children with cystic fibrosis, diabetes, and mental retardation. *Journal of Pediatric Psychology* **17**, 327–43.

Weiss M. J. (2002) Hardiness and social support as predictors of stress in mothers of typical children, children with autism, and children with mental retardation. *Autism* **6**, 115–30.

Witt W. P., Riley A. W. & Coiro M. J. (2003) Childhood functional status, family stressors, and psychosocial adjustment among school-aged children with disabilities in the United States. *Archives of Pediatrics and Adolescent Medicine* **157**, 687–95.

World Bank (2020) World Bank Country and Lending Groups. Country classification. Available at: https://datahelpdesk.worldbank.org/knowledgebase/articles/906519-world-bank-country-and-lending-groups

A: Appendix 1: search strategy

Search methods for identification of studies

We searched the following electronic databases for all available years for English language papers: the Cumulative Index to Nursing and Allied Health Literature (CINAHL), Excerpta Medica dataBASE (EMBASE), Medical Literature Analysis and Retrieval System Online (MEDLINE) and Psychological Information Database (PsycINFO).

Electronic searches

We used the following Ovid MEDLINE search strategy and adapted it as appropriate for other databases:

1 ((mental*) adj3 (handicap* or retard*)).tw.
2 ((disabilit*) adj3 (intellectual)).tw.
3 or/1–2
4 depress*.mp.
5 (mental disorder* or anxi* or anxiety disorder* or (bipolar and related disorder*) or behaviour disorder* or behavior disorder* or delirium or dissociative disorder* or emotional disorder* or mental instability or mood disorder* or affective disorder* or neurosis or personality disorder* or psychosis or psychosomatic disorder* or psychiatric disorder* or stress* or stress related disorder* or stress-related disorder* or paranoid disorder* or psychotic disorder*).mp.
6 psychopatholog*.tw.
7 mental health.tw.
8 mental ill-health.tw.
9 mental ill health.tw.
10 (mental adj2 (disorder* or problem* or condition*)).tw.
11 (well-being or wellbeing).tw.
12 or/4–11
13 (mothers or parent).mp.
14 mother*.tw.
15 maternal.tw.
16 (caregiver* or care-giver*).mp.
17 carer*.tw.
18 (mum* or mom*).tw.
19 parent*.tw.
20 or/13–19
21 3 and 12 and 20
22 Humans/
23 21 and 22

Searching other resources

We checked the reference list of all relevant studies in order to find additional publications which may have been missed by our electronic searches (e.g. unpublished or in-press citations).

B: Appendix 2: quality assessment scoring for individual CASP items

CASP item[†]	Cummings *et al*. 1966	Miller and Keirn 1978	Harris and McHale 1989	Walker *et al*. 1989	Walker *et al*. 1992
1. Did the study address a clearly focused issue?	Yes	Yes	Yes	Yes	Yes
2. Was the cohort recruited in an acceptable way?	Yes	Yes	Yes	Yes	Yes
3. Was the exposure accurately measured to minimise bias?	Yes	Yes	Cannot tell	Yes	Yes
4. Was the outcome accurately measured to minimise bias?	Yes	Cannot tell	Yes	Yes	Yes
5. (a) Have the authors identified all important confounding factors?	Yes	No	Cannot tell	Yes	No
5. (b) Have they taken account of the confounding factors in the design and/or analysis?	Yes	No	Cannot tell	Yes	No
6. (a) Was the follow up of subjects complete enough?	Yes	Yes	Yes	Yes	Yes
6. (b) Was the follow up of subjects long enough?	Yes	Yes	Yes	Yes	Yes
7. What are the results of this study?	Yes	Yes	Yes	Yes	Yes
8. How precise are the results?	Yes	Yes	Yes	Yes	Yes
9. Do you believe the results?	Yes	Yes	Yes	Yes	Cannot tell
10. Can the results be applied to the local population?	Yes	Yes	Yes	Yes	Yes
11. Do the results of this study fit with other available evidence?	Yes	Yes	Yes	Cannot tell	No
12. What are the implications of this study for practice?	Yes	No	Yes	Yes	Yes
Total score	A	C2.1	B2	B1	C2.1

[†]Possible scorings for individual items: yes (*low risk*), cannot tell (*unknown risk*), no (*high risk*).

CASP, Critical Appraisal Skills Programme Checklist.

Rating A = low risk of bias for all 12 items.

Rating B*x* = uncertain risk of bias for *x* items, low risk of bias in all other items.

Rating C*y*,*x* = high risk of bias in *y* items, uncertain risk of bias in *x* items, low risk of bias in all other items.

CASP item[†]	Andersson 1993	Ong *et al*. 1999	Olsson and Hwang 2001	Rimmerman and Muraver 2001	Olsson and Hwang 2002	Weiss 2002
1. Did the study address a clearly focused issue?	Yes	Yes	Yes	Yes	Yes	Yes
2. Was the cohort recruited in an acceptable way?	Yes	Yes	Yes	Yes	Yes	Yes
3. Was the exposure accurately measured to minimise bias?	Yes	Yes	Yes	Yes	Yes	Yes
4. Was the outcome accurately measured to minimise bias?	Yes	Yes	Yes	Yes	Yes	Yes
5. (a) Have the authors identified all important confounding factors?	No	Cannot tell	Yes	Cannot tell	Yes	Yes
5. (b) Have they taken account of the confounding factors in the design and/or analysis?	No	Cannot tell	Yes	Cannot tell	Yes	Yes
6. (a) Was the follow up of subjects complete enough?	Yes	Yes	Yes	Yes	Yes	Yes
6. (b) Was the follow up of subjects long enough?	Yes	Yes	Yes	Yes	Yes	Yes
7. What are the results of this study?	Yes	Yes	Yes	Yes	Yes	Yes
8. How precise are the results?	No	Yes	Yes	Yes	Yes	Yes
9. Do you believe the results?	Yes	Yes	Yes	Yes	Yes	Yes
10. Can the results be applied to the local population?	Yes	Yes	Yes	Yes	Yes	Yes
11. Do the results of this study fit with other available evidence?	Yes	Yes	Yes	Yes	Yes	Yes
12. What are the implications of this study for practice?	Yes	Yes	Yes	Yes	Cannot tell	Yes
Total score	C2	B1	A	B1	B1	A

[†]Possible scorings for individual items: yes (*low risk*), cannot tell (*unknown risk*), no (*high risk*).

CASP, Critical Appraisal Skills Programme Checklist.

Rating A = low risk of bias for all 12 items.

Rating B*x* = uncertain risk of bias for *x* items, low risk of bias in all other items.

Rating C*y*,*x* = high risk of bias in *y* items, uncertain risk of bias in *x* items, low risk of bias in all other items.

CASP item[†]	Emerson 2003	Eisenhower *et al*. 2005	Lenhard *et al*. 2005	Abasiubong *et al*. 2006	Emerson *et al*. 2006
1. Did the study address a clearly focused issue?	Yes	Yes	Yes	Yes	Yes
2. Was the cohort recruited in an acceptable way?	Yes	Yes	Yes	Yes	Yes
3. Was the exposure accurately measured to minimise bias?	Cannot tell	Cannot tell	Cannot tell	Yes	Cannot tell
4. Was the outcome accurately measured to minimise bias?	Yes	Yes	Cannot tell	Yes	Cannot tell
5. (a) Have the authors identified all important confounding factors?	No	Yes	Yes	Cannot tell	Yes
5. (b) Have they taken account of the confounding factors in the design and/or analysis?	No	Yes	Cannot tell	Cannot tell	Yes
6. (a) Was the follow up of subjects complete enough?	Yes	Yes	Yes	Yes	Yes
6. (b) Was the follow up of subjects long enough?	Yes	Yes	Yes	Yes	Yes
7. What are the results of this study?	Yes	Yes	Yes	Yes	Yes
8. How precise are the results?	No	Yes	No	No	No
9. Do you believe the results?	Yes	Yes	Yes	Yes	Yes
10. Can the results be applied to the local population?	Yes	Yes	Yes	Yes	Yes
11. Do the results of this study fit with other available evidence?	Yes	Yes	Yes	Yes	Yes
12. What are the implications of this study for practice?	Yes	Yes	Yes	Yes	No
Total score	C2.1	B1	C1.3	C1.1	C2.2

[†]Possible scorings for individual items: yes (*low risk*), cannot tell (*unknown risk*), no (*high risk*).

CASP, Critical Appraisal Skills Programme Checklist.

Rating A = low risk of bias for all 12 items.

Rating B*x* = uncertain risk of bias for *x* items, low risk of bias in all other items.

Rating C*y*,*x* = high risk of bias in *y* items, uncertain risk of bias in *x* items, low risk of bias in all other items.

CASP item[†]	Olsson and Hwang 2006	Mugno *et al.* 2007	Neece and Baker 2008	Olsson and Hwang 2008	Gupta and Kaur 2010
1. Did the study address a clearly focused issue?	Yes	Yes	Yes	Yes	Yes
2. Was the cohort recruited in an acceptable way?	Yes	Yes	Yes	Yes	Yes
3. Was the exposure accurately measured to minimise bias?	Yes	Yes	Yes	Cannot tell	Yes
4. Was the outcome accurately measured to minimise bias?	Yes	Yes	Yes	Yes	Yes
5. (a) Have the authors identified all important confounding factors?	Cannot tell	Yes	Yes	Yes	No
5. (b) Have they taken account of the confounding factors in the design and/or analysis?	Cannot tell	Yes	Yes	Yes	No
6. (a) Was the follow up of subjects complete enough?	Yes	Yes	Yes	Yes	Yes
6. (b) Was the follow up of subjects long enough?	Yes	Yes	Yes	Yes	Yes
7. What are the results of this study?	Yes	Yes	Yes	Yes	Yes
8. How precise are the results?	Yes	Yes	Cannot tell	Yes	No
9. Do you believe the results?	Yes	Yes	Yes	Yes	Yes
10. Can the results be applied to the local population?	Yes	Yes	Yes	Yes	Yes
11. Do the results of this study fit with other available evidence?	Yes	Yes	Yes	Yes	Yes
12. What are the implications of this study for practice?	Cannot tell	Yes	Yes	Cannot tell	No
Total score	B2	A	B1	B2	C3

[†]Possible scorings for individual items: yes (*low risk*), cannot tell (*unknown risk*), no (*high risk*).

CASP, Critical Appraisal Skills Programme Checklist.

Rating A = low risk of bias for all 12 items.

Rating B*x* = uncertain risk of bias for *x* items, low risk of bias in all other items.

Rating C*y*,*x* = high risk of bias in *y* items, uncertain risk of bias in *x* items, low risk of bias in all other items.

CASP item[†]	Mirsaleh *et al.* 2011	Totsika *et al.* 2011a	Totsika *et al.* 2011b	Kilic *et al.* 2013	Norlin and Broberg 2013
1. Did the study address a clearly focused issue?	Yes	Yes	Yes	Yes	Yes
2. Was the cohort recruited in an acceptable way?	Yes	Yes	Yes	Yes	Yes
3. Was the exposure accurately measured to minimise bias?	Yes	Yes	Cannot tell	Cannot tell	Yes
4. Was the outcome accurately measured to minimise bias?	Yes	Yes	Yes	Yes	Yes
5. (a) Have the authors identified all important confounding factors?	No	Cannot tell	Cannot tell	No	Yes
5. (b) Have they taken account of the confounding factors in the design and/or analysis?	No	Cannot tell	Cannot tell	No	Yes
6. (a) Was the follow up of subjects complete enough?	Yes	Yes	Yes	Yes	Yes
6. (b) Was the follow up of subjects long enough?	Yes	Yes	Yes	Yes	Yes
7. What are the results of this study?	Yes	Yes	Yes	Yes	Yes
8. How precise are the results?	Yes	Yes	Yes	Yes	Yes
9. Do you believe the results?	Yes	Yes	Yes	Yes	Yes
10. Can the results be applied to the local population?	Yes	Yes	Yes	Yes	Yes
11. Do the results of this study fit with other available evidence?	Yes	Yes	Yes	Yes	Yes
12. What are the implications of this study for practice?	No	Yes	Yes	Yes	Yes
Total score	C2	B1	B2	C1.1	A

[†]Possible scorings for individual items: yes (*low risk*), cannot tell (*unknown risk*), no (*high risk*).

CASP, Critical Appraisal Skills Programme Checklist.

Rating A = low risk of bias for all 12 items.

Rating B*x* = uncertain risk of bias for *x* items, low risk of bias in all other items.

Rating C*y*,*x* = high risk of bias in *y* items, uncertain risk of bias in *x* items, low risk of bias in all other items.

CASP item[†]	Fairthorne *et al.* 2015	Long *et al.* 2015	Faithorne *et al.* 2016	AlAnsari and Jahrami 2018	Hosseininik *et al.* 2018	Blacher and Baker 2019
1. Did the study address a clearly focused issue?	Yes	Yes	Yes	Yes	Yes	Yes
2. Was the cohort recruited in an acceptable way?	Yes	Yes	Yes	No	Yes	Yes
3. Was the exposure accurately measured to minimise bias?	Yes	Yes	Yes	Yes	Cannot tell	Yes
4. Was the outcome accurately measured to minimise bias?	Yes	Yes	Yes	Yes	Yes	Yes
5. (a) Have the authors identified all important confounding factors?	Yes	Yes	Yes	Yes	No	Yes
5. (b) Have they taken account of the confounding factors in the design and/or analysis?	Yes	Yes	Yes	Cannot tell	No	Yes
6. (a) Was the follow up of subjects complete enough?	Yes	Yes	Yes	Yes	Yes	Yes
6. (b) Was the follow up of subjects long enough?	Yes	Yes	Yes	Yes	Yes	Yes
7. What are the results of this study?	Yes	Yes	Yes	Yes	No	Yes
8. How precise are the results?	Yes	Cannot tell	Yes	Yes	No	Yes
9. Do you believe the results?	Yes	Yes	Yes	Yes	No	Yes
10. Can the results be applied to the local population?	Yes	Yes	Yes	Yes	Cannot tell	Yes
11. Do the results of this study fit with other available evidence?	Yes	Yes	Yes	Yes	Yes	Yes
12. What are the implications of this study for practice?	Yes	Yes	Yes	No	No	Yes
Total score	A	B1	A	C2.1	C5.2	A

[†]Possible scorings for individual items: yes (*low risk*), cannot tell (*unknown risk*), no (*high risk*).

Rating A = low risk of bias for all 12 items.

Rating B*x* = uncertain risk of bias for *x* items, low risk of bias in all other items.

Rating C*y*,*x* = high risk of bias in *y* items, uncertain risk of bias in *x* items, low risk of bias in all other items.

Accepted 20 February 2021